SCIENTIFIC ARTICLE

An underrated differential diagnosis for subcutaneous lumps: 26 cases of subcutaneous Dirofilariasis

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Abstract

Introduction

Even though subcutaneous dirofilariasis [worm granuloma] is an emerging zoonosis, it is not commonplace to consider it as a differential diagnosis for a subcutaneous lump even in the endemic zones. Moreover, the available literature lacks attempts to provide a unified clinical profile for it to be considered as a clinical entity.

Methods

We have retrospectively analyzed patients who were diagnosed with subcutaneous dirofilariasis at a Base Hospital in the Uva Province of Sri Lanka from February 2018 to March 2020. We evaluated patient demographics, symptomatology, clinical signs, ultrasonographic and histopathological features.

Results

Twenty-six patients with a mean age of 15.6 years [range 1 – 67 years] were analyzed. The locations of the lumps were highly variable, the commonest being the abdominal wall [34.6%]. Most [84.6%] were asymptomatic. The average diameter was 14.2mm [range 8 – 25mm]. The majority were neither tender nor warm, firm, and had ill-defined margins and a smooth surface without fluctuation, transillumination, or slipping sign. All were in the subcutaneous plane often attached to the deep fascia but without attachment to the skin. Blood investigations were unremarkable with a normal ESR and eosinophil count. The ultrasonographic finding was an echogenic tubular structure within a hypoechoic lesion. Histopathology demonstrated a worm surrounded by granulomatous inflammatory infiltrates with eosinophils and lymphocytes.

Conclusion

We emphasize the importance of regarding subcutaneous dirofilariasis as a differential diagnosis for subcutaneous lumps, especially in the highly prevalent geographical zones to minimize 'incidental diagnosis'.

Introduction

Human dirofilariasis is an emerging zoonotic infestation producing parasitic granuloma typically with subcutaneous, pulmonary, and ocular involvement [1]. Nematodes of genus Dirofilaria commonly infect wild and domestic animals, particularly canines. Out of about 40 recognized species of Dirofilaria, few of them including D. immitis, D. repens, D. striata, D. tenuis, D. ursi and D. spectans infect humans accidentally [1, 2]. Mosquitos of genera Anopheles, Aedia, Culex and Mansonia act as vectors in the transmission of the disease. In addition, fleas and ticks are also recognized as vectors. [2]

The condition is widespread involving both temperate and tropical climates around the world. Southern and Central Asia, Southern and Eastern Europe have been recognized as the highest endemic zones. Among the Asian countries, Sri Lanka is the most endemic country followed by Malaysia [1, 3]. D. repens is the commonest reported nematode responsible for subcutaneous dirofilariasis in South East Asia including Sri Lanka. Condition is manifested as subcutaneous lumps commonly affecting the exposed areas of the body. e. g. Face, upper and lower limbs. The preference for the exposed areas is believed to be due to the tendency of the granuloma to locate at the sites of mosquito bites [4, 6].

Even though there is a rising trend of incidence, it is not commonplace to consider parasitic granuloma as a differential for a subcutaneous lump among the other classic differential diagnoses. The available world literature describes parasitic and vector biology, the geographic distribution of the disease as well a retrospective series of unusual subcutaneous lumps which were diagnosed as dirofilariasis by radiology and histology. We could not observe any attempt to provide a unified clinical profile for subcutaneous dirofilariasis to be considered in the clinical diagnosis rather than being an 'unusual lump'.

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Methods

We have retrospectively analyzed patients who were diagnosed with subcutaneous dirofilariasis at Base Hospital Mahiyangana from February 2018 to March 2020 after obtaining approval from the institutional ethical review board and administration. The data were retrieved from the patients during initial and follow-up clinic visits after obtaining written informed consent.

We evaluated the data on patient demographics, clinical signs, symptomatology, ultrasonographic and histopathological features. All the patients underwent an ultrasonographic assessment on clinical suspicion and later underwent excision biopsy for histological evaluation if ultrasonographic features were compatible with the clinical diagnosis.

Results

A total of 26 patients had been evaluated over 2 years. The mean age at presentation was 15.6 years [range 1 – 67 years] and the majority [61.5%] belonged to the paediatric age group. The distribution of the location of the lumps was highly variable, most common being the abdominal wall [34.6%] followed by lower limbs [19.2%]. Two patients had lumps in unusual locations including buccal mucosa and breast.

Symptoms and signs were highly variable among the reported cases. In the paediatric group, most of the lumps were noticed incidentally by their parents during bathing. Adults were asymptomatic except for a few patients with mild pain and itching. The majority of the lumps were oval shaped with an average diameter of 14.2 mm [range 8 – 25 mm]. Skin changes were minimal except for a lump that mimicked a keloid. The majority were non-tender or mildly tender, not warm, firm in consistency and having ill-defined margins and a smooth surface without fluctuation, transillumination or slipping sign. All of them were in the subcutaneous [submucosal in the case of buccal lump] plane often attached to deep fascia but without attachment to the skin.

Blood investigations were found to be unremarkable with a normal ESR, white blood cell count and eosinophil count. The general ultrasonographic finding was an echogenic tubular structure within a hypoechoic lesion. The characteristic histological finding was the demonstration of a worm surrounded by granulomatous inflammatory infiltrates with eosinophils, lymphocytes, plasma cells and histiocytes.

Discussion

Following the available literature, we have noticed a wide age range of affected patients [1 – 67 years] depicting that a person of any age can acquire the condition. However, there is some predilection towards the pediatric age group.

The location of subcutaneous dirofilariasis is highly variable with the possibility of producing lumps in almost any location of the body. Contrary to the previously published series, we observed the commonest location as abdominal wall [34.6%] followed by lower limbs rather than on exposed areas of the body including the face [4, 6]. Sizes of the lumps are variable in the literature, many approximating 15 – 20mm [1] which is compatible with the average diameter of our series [14.2mm].

Diagnostic challenges were noticed with the signs related to the lumps when considering the other classic differential diagnoses on the particular location. However, examination with attention to detail revealed certain differences from the other common differential diagnoses.

Skin changes associated with the lumps were minimal except in one case who presented with a breast lump [figure 1a] that mimicked a keloid but without a history of local tissue damage. The majority mimicked implantation dermoid due to the subcutaneous location, firm consistency, and lack of transillumination. However, ill-defined margins, lack of fluctuation and deep attachment allowed the differentiation from a dermoid. The main differentiating features from sebaceous cyst were lack of skin attachment, absence of punctum, ill-defined margins and non-fluctuant nature. Lipoma was excluded from the differential diagnosis as the lumps were firm, non-fluctuant and absent of slipping sign. Few lumps that were located on the hand and foot could be differentiated from a ganglion by the absence of fluctuation and transillumination.

Two other patients with lumps located in less frequent sites posed issues with clinical diagnosis. A 3-year-old boy presenting with an anterior neck lump just off the midline and having attachment to the investing layer of deep fascia

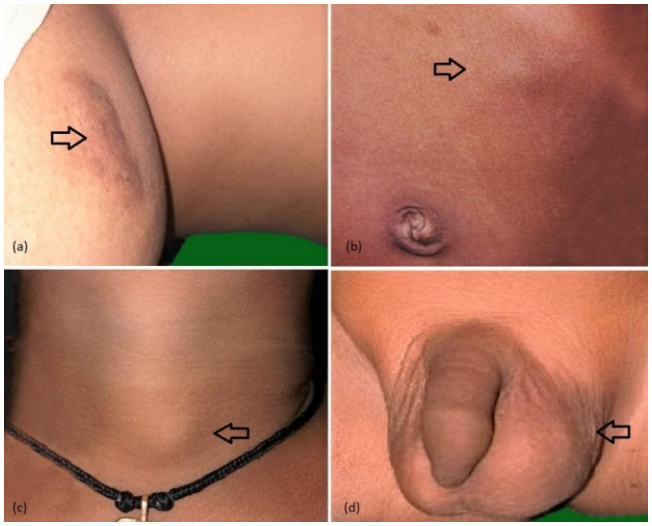

Figure 1. various location sof worm granuloma (a) right breast (b) abdominal wall (c) anterior neck (d) left hemiscrotum

resembled a thyroid nodule [figure 1c]. It was differentiated from a thyroglossal cyst by its lack of mobility with tongue protrusion. A 5-year-old boy with a lump over the left hemi scrotum which mimicked an epididymal cyst but lacked a typical 'Chinese lantern appearance' with transillumination [figure 1d].

Blood eosinophilia and elevation of inflammatory markers are rarely elevated, and they are of limited value in the diagnosis of subcutaneous dirofilariasis [3, 4]. Laboratory assessment of our series was unremarkably complying with the previous studies. Ultrasound scan aided the clinical diagnosis by demonstrating an echogenic tubular structure within a hypoechoic lesion in almost all the cases [figure 2].

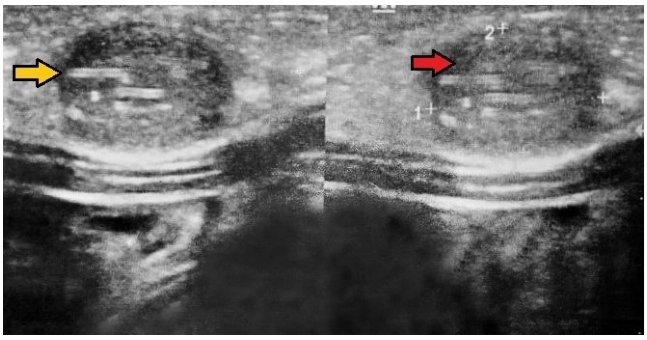

Figure 2. Ultrasonography showing an echogenic tubular structure (yellow arrow head) within hypoechoic lesion (red arrow head) in the subcutaneous tissue plane.

Since there is no migration into the bloodstream, the anthelminthic treatment is less effective, so the treatment of choice is surgical excision[7, 8]. However, treatment with a course of Ivermectin and Diethylcarbamazine [DEC] is in use if secondary lesions developed to prevent further surgical excisions [8]. However, we did not observe such recurrences in our participants during the limited follow-up.

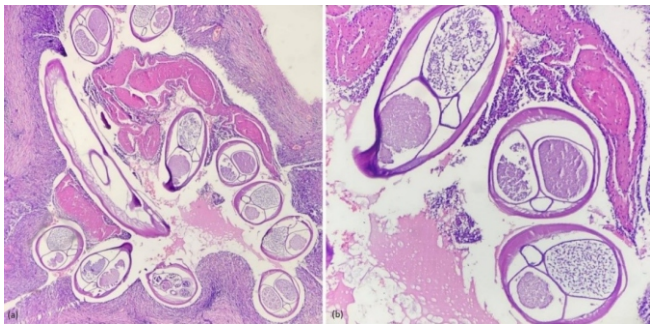

Figure 2. Tangenital and cross sections of dirofilaria worm showing well developed multi layered cuticle, alimentary canal and genital tube. The worm is surrounded by dense inflammatory infiltrate with fleets of fleets of eosinophils, lymphocytes, plasma cells and histiocytes. Haemotoxylin and eosin; magnification (a) X 40 (b) X 100.

The histological evaluation confirmed the diagnosis, demonstrating the worm surrounding granulomatous inflammation with fleets of eosinophils, lymphocytes, plasma cells and histiocytes [figure 3].

Conclusion

This article adds to the increasing body of evidence of dirofilariasis in Sri Lanka. We emphasize the importance of subcutaneous dirofilariasis in the differential diagnosis for subcutaneous lumps, especially in the highly prevalent geographical zones that minimize the diagnostic errors.

In addition, there is an increasing trend of the paediatric population being affected. Standard hygiene and vector control are to be considered to overcome this parasitic infestation.

All authors disclose no conflict of interest. The study was conducted in accordance with the ethical standards of the relevant institutional or national ethics committee and the Helsinki Declaration of 1975, as revised in 2000.

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