Cheek swelling: An unusual presentation of filariasis

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Filariasis and its consequences are a major health problem in tropical countries like India\(^1\). *Wuchereria bancrofti, Brugia malayi* and *Brugia timori* are the three nematodes responsible for the filariasis. Of these, only *W. bancrofti* and *B. malayi* are found in India. The disease has a predilection for lower limbs, spermatic cord and epididymis\(^2\). The breast\(^3\), thyroid\(^4\), body fluids\(^5\) and skin\(^6\) are unusual sites for filariasis. Oral or perioral involvement is even rarer\(^7\)-\(^8\). An unusual presentation as a cheek swelling prompted us to report the present case.

**Case report**

A 35-yr-old lady presented to the Otolaryngorhino-logy outpatient department with the complaint of a right cheek swelling since eight days, which was gradually increasing in size. The swelling was diffuse, euthermic and non tender. The skin overlying was devoid of any specific lesion (Fig. 1). She was afebrile with no lymphadenopathy or organomegaly. Examination of her complete blood count (CBC) revealed absolute eosinophilia (1080/mm\(^3\)) but she was negative for any parasitic form in the peripheral blood smear. With this clinical presentation, she was advised ultrasonography (USG) and fine needle aspiration cytology (FNAC) of the swelling. The USG of swelling showed the evidence of cold abscess.

FNAC samples yielded creamy material. Haematoxyline and eosin (H & E) stained smears of aspirate exhibited sheathed dead microfilarial worm. The background showed a granulomatous reaction comprising of epithelioid histiocytes, multinucleated giant cells and many eosinophils (Fig. 2). Diagnosis of granulomatous reaction to dead microfilaria was offered. The excisional biopsy of the swelling was performed. The H & E stained section demonstrated fibro-fatty tissue, within which the hyalinised filarial worm with granulomatous reaction and dense eosinophillic infiltrate was noted. Within the worm occasional microfilaria was identified, probably as a part of gravid uterus of the worm (Fig. 3). Only because of the morphologically identifiable microfilaria in the cytology smears, we could guess the hyalinised worm as filarial worm which was near impossible otherwise. As only dead and degenerated forms of worm were found,
further typing of worm was not possible even after many attempts. The patient was advised diethylcarbamazine for 15 days. After completion of treatment, she is asymptomatic and her CBC is within normal limits.

DISCUSSION

Filariasis is common in tropical and subtropical areas of Africa, Asia, South and Central America. It is endemic in some parts of India. The causative organisms are *W. bancrofti*, *B. malayi* and *B. timori*. The nematodes like *Loa loa*, *Onocerca volvulus*, and *Mansonella* are known to cause subcutaneous filariasis7. In India, the most widespread filariasis infection is due to *W. bancrofti* (98%)9. The variants of these two species have been featured by differences in the natural host range, microfilarial periodicity, and vector susceptibility. *W. bancrofti* and *B. malayi* are nocturnally periodic.

The disease caused by filarial worm can be acute with fever and chills while manifestations of chronic disease include lymphangitis, lymphadenitis, hydrocele and elephantiasis2. Incidental detection of filarial organism, mostly microfilarial form, has been reported in cytological smears from almost all body parts. Adult worms are found in regional lymphatics, while microfilariae circulate in the blood. As the parasite circulate in the lymphatic and vascular systems they can cause lymphovascular obstruction resulting into extravasations of blood and release of microfilariae in the subcutaneous plane10. Patient may present as a subcutaneous nodular swelling in absence of any other symptom. Oral or perioral involvement of the filarial nematode is rare11.

In the present case, patient appeared to be an asymptomatic carrier with larvae present in microvasculature. A rupture of vessels might have led to haemorrhage and release of microfilariae into the subcutaneous tissue. Surrounding inflammatory reaction probably led to the development of a well-defined cystic swelling containing multiple microfilariae, which subsequently developed into adult filarial worms.

In the recent past, diagnosis of filariasis rested on demonstration of the microfilariae in blood. The microfilariae of *W. bancrofti* mainly exhibit nocturnal periodicity and peripheral blood samples need to be taken at night, between 2200 and 1400 hrs2. Recently, FNAC has proved to be an important tool in diagnosis of filarial diseases as it is used as a confirmatory diagnostic tool in suspected cases of filariasis of testis, epididymis, lymph node, thyroid and subcutaneous nodules12.

On cytological examination, microfilariae can be evident with or without adult worms along with eosinophils, neutrophils, and mononuclear cells. The presence of epithelioid cell granulomas and giant cells are other usual findings in filariasis. This can create a diagnostic dilemma and needs to be differentiated from other granulomatous diseases such as tuberculosis and leprosy, because of common cytomorphological features. Presence of worm–adult or larval form, and the negative 20 and 5% Ziehl-Neelsen stains pinpoints the diagnosis.

Filariasis may present in an unusual fashion so, careful screening of cytology smears, even when filariasis is not suspected, as in this case, is medically important for diagnosis and treatment of the patients, especially in endemic areas.

REFERENCES


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**CORRIGENDUM**

**Conflict of first reporting**

This has reference to the following article published in the June issue (2015) of the journal:


Readers may note that a similar study (with contemporaneous sampling period but different approach) was published earlier by another research group in a Spanish journal (indicated below), reporting about the continuity of reproductive cycle of *Aedes albopictus* during winter season in Europe.