Case Report
Lymphadenovarix of the Head–Neck region—A Rare Presentation of Bancroftian Filariasis

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Summary
Cystic swellings of the neck in children have limited differential diagnoses, often either lymphatic or vascular malformations. Other cystic inflammations can be the result of tuberculous abscesses, suppurated lymph nodes and actinomycosis. Microfilaria causing lymphadenovarix of head–neck region has not yet been described in the literature. A 10-year-old Indian boy presented with an asymptomatic cystic neck mass of 8 months duration. Aspiration of the swelling demonstrated numerous Wuchereria bancrofti microfilaria and the patient responded well to 6 weeks of daily anti-filarial treatment using diethylcarbamazine citrate (6mg kg\(^{-1}\) day\(^{-1}\)). This appears to be the first report of microfilariae-associated lymphadenovarix of head–neck region. Though rare, filariasis should be considered as a differential diagnosis for aberrant swellings where lymphatic filarids are endemic.

Key words: Posterior triangle, Microfilaria, Wuchereria bancrofti.

Introduction
Lymphatic filariasis, affects over 120 million people in 80 countries. More than 1.1 billion people live in areas where there is a risk of infection [1]. Filariasis and its consequences are a major health problem in tropical countries, including the Indian subcontinent. Majority of the infected individuals in filariasis endemic communities are asymptomatic. Currently there may be up to 31 million microfilaraemics, 23 million cases of symptomatic filariasis and about 473 million individuals potentially at risk of infection in India [2]. The State of Bihar has the highest endemicity (over 17%) followed by Kerala (15.7%) and Uttar Pradesh (14.6%) [2]. Despite its high incidence, it is unusual to find microfilaria in fine needle aspiration cytology (FNAC) smears. Very few cases have been diagnosed by fine needle aspiration cytology (FNAC) from different sites in the body like breast, thyroid, lymph node, liver, lungs, skin nodules, bone marrow and body fluids [3–8]; but microfilaria from lymphadenovarix of head-neck region has never been reported in the literature until now.

Case report
A 10-year-old Indian boy presented with an asymptomatic swelling in the right posterior triangle of the neck for an 8 month duration. On inspection the swelling was, 3 \(\times\) 3 cm, well defined, cystic, non-tender and compressible. The temperature of swelling was not raised. It appeared to arise from deep to the anterior border of trapezius muscle. It was non-pulsatile and was not associated with any cough impulse or bruit. It became less prominent when the patient was asked to shrug his shoulders against resistance, and remained in the same position when the sternomastoid muscle of the same side was made to contract (Fig. 1). Ultrasonography of swelling confirmed the findings of a cystic consistency and extension into the anterior fibers of trapezius muscle (Fig. 2). There was no loss of function and patient had no other symptoms or signs attributable to filariasis.

A fine needle aspiration (FNA) was performed. The aspirate was sero-sanguinous smeared on a slide, air dried and stained with Giemsa. The cytologic
examination revealed four sheathed microfilaria (measuring 270–300 × 8–10 μm) with granules (nuclei) not extending up to the tip of tail. A caudal space of 5–15 μm was seen at the posterior end, confirming it to be *Wuchereria bancrofti* (Fig. 3). Cells like eosinophils, neutrophils, histiocytes and multinucleated giant cells were also seen in the smear. Routine hematological investigations were normal and the peripheral blood film failed to reveal eosinophilia or presence of any microfilaria. The aspirate was also cultured for bacteria but was found to be sterile.

The patient was prescribed diethylcarbamazine (DEC, 6 mg kg⁻¹ day⁻¹) for 2 weeks and was asked to come for follow-up. There was slight reduction in the size of swelling after 2 weeks. Treatment was continued for two more weeks and the patient was asked to come again. On follow-up there was some more reduction in the size of swelling. The standard recommendation for using DEC is 3 weeks but in view of the slow, but good response, the treatment was continued for 6 weeks. After 6 weeks of DEC treatment, the swelling had almost disappeared (Fig. 4). Several blood smears (nocturnal and diurnal) were done on follow-up, but yielded no microfilaria. Membrane filtration concentration technique and DEC provocation test for detection of microfilaria were not used.

**Discussion**

Filariasis is a major public health problem in India. It is mainly a disease of adults and is more common in men [9, 10] with most infections caused by *W. bancrofti*. The definitive diagnosis of filariasis depends on demonstration of the parasite either in peripheral blood or in the aspirate. Microfilaria or
worm fragments may be seen with FNAC [11]. Indirect haemagglutination, ELISA, immunoassays and polymerase chain reaction (PCR) are also helpful in making the diagnosis [12, 13]. If microfilariae cannot be found, DEC challenge test needs to be done to demonstrate microfilaria. The diagnosis must be made on clinical grounds by the exclusion of other causes.

Clinical features and pathology depend on the sites occupied by developing and mature worms, the number of worms present, duration of infection and the immune response of the host, especially to damaged and dead worms. The disease has a wide spectrum of presentation from asymptomatic lymphatic filariasis, acute lymphatic filariasis and chronic lymphatic filariasis. Many patients remain asymptomatic despite the presence of a microfilaraemia in peripheral blood.

Chronic lymphatic filariasis is frequently found and may be the only manifestation of filariasis, hydrocele being the most common feature. Other manifestations of chronic disease are elephantiasis of the limbs and chyluria.

The diagnosis of a filarial infection can also been made by detecting microfilariae on microscopic examination of fine needle aspirates from lymph nodes [3, 14]. FNAC of breast mass, thyroid mass, hydrocele fluid, pericardial fluid, pleural fluid, ascitic fluid and cytology of cervicovaginal smears, bronchial aspirates, urine, nipple secretion, bone marrow and joint fluid aspirates have also been reported to yield microfilariae [15, 16]. Moreover, in these patients the peripheral smears rarely revealed microfilaraemia or eosinophilia [15, 16] as was also seen in our case.

The present case did not have any clinical evidence of filariasis and there was no microfilaraemia, i.e. the patient had occult filariasis. In occult filariasis, microfilariae are found in affected tissues but not in peripheral blood. This can be seen in endemic areas like ours where filariasis can exist without microfilaraemia or microfilaraemia may be extremely transient and hence overlooked [17].

There is no satisfactory treatment for filariasis. Diethylcarbamazine 6 mg kg\(^{-1}\) day\(^{-1}\) for 2–3 weeks or Ivermectin single dose of 200–400 µg kg\(^{-1}\) can be used, but it should be repeated every 6–12 months. Single dose of Ivermectin at 200–400 µg kg\(^{-1}\) plus Albendazole 400 mg are reported to be more effective in some studies [14]. In our case, we used DEC alone for treatment. The safety and efficacy against lymphatic filariasis of the drug combination DEC and albendazole was studied in a randomized controlled trial in India that showed the drug combination to be as safe as DEC alone, and that both drugs were adequately absorbed. However, in this trial there was no evidence of greater efficacy of the combination at 12 months follow-up [18].

In conclusion, filariasis should be considered as a differential diagnosis for aberrant swellings where lymphatic filarids are endemic.

References