

CASE REPORT

Facial wrigglies: live extralymphatic filarial infestation in subcutaneous tissues of the head and neck

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ABSTRACT. We report a rare case of a 32-year-old male with live extralymphatic filarial infestation presenting as a facial subcutaneous soft-tissue swelling. To the best of our knowledge these imaging findings have not been previously reported in the head and neck region in the existing English language literature. Real-time high-resolution ultrasonography revealed a solitary well-defined subcutaneous cystic lesion over the right zygomatic arch. It showed multiple linear, echogenic, undulating structures exhibiting a persistent twirling motion during the examination. This typical ultrasonographic appearance was consistent with the filarial dance sign (FDS) of live adult filarial worms. Subsequent MRI confirmed the cystic and solitary nature of the lesion. Complete excision of the cyst was performed, which revealed intracystic straw-coloured fluid and multiple white-coloured adult worms within the lesion. Histopathological examination confirmed multiple adult filarial worms with surrounding reactive inflammatory changes. In an endemic region, identification of the FDS in any normal anatomical structure or abnormal swelling, however remote or unusual the location within the body, should strongly suggest the diagnosis of live active filarial infestation. In view of the increasing migratory trends in the global population, it is imperative for radiologists in all countries to be aware of the typical imaging findings of this disease to arrive at the correct diagnosis.

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Filariasis is a common public health problem in South East Asia, parts of Africa and the western Pacific region and is widely distributed through the tropical and subtropical regions [1, 2]. There are approximately 120 million people infected in these regions and approximately 75 million do not show the clinical manifestations of this disease [3]. Filariasis in the Asian subcontinent is caused by two closely related nematode worms, *Wuchereria bancrofti* and *Brugia malayi* [4]. The disease mainly involves the lymphatic system of the body. The most frequently involved parts of the lymphatic system are the lower limbs, retroperitoneal tissues, spermatic cord, epididymis and mammary glands [5, 6].

We report an unusual case of extralymphatic filarial infestation in a solitary cystic swelling located in the facial subcutaneous soft tissue. This diagnosis was possible because of the typical sonographic appearance, the filarial dance sign (FDS), of live adult filarial worms described by Amaral et al [7] and Suresh et al [8]. To the best of our knowledge such a finding has never been documented within facial subcutaneous tissues in the existing English language literature.

Case report

A 32-year-old male patient was referred for ultrasound evaluation of a painless soft-tissue swelling over the right zygomatic arch (Figure 1). The swelling had gradually increased in size over a period of 3 months. Real-time sonography was performed on a Philips HDI 5000 SonoCT ultrasound scanner (Philips Healthcare, Andover, MA) using a 12 Mhz, high-frequency transducer. It showed a solitary, well-defined lesion measuring 1.7 × 0.6 × 2.5 cm (anteroposterior, transverse and cranio-caudal dimensions) in the facial subcutaneous soft tissues. The lesion was predominantly cystic with a thick capsule and contained multiple linear echogenic structures (Figure 2). These linear echogenic structures showed a persistent twirling motion on real-time sonography which was video recorded for documentation purposes. The ultrasonographic appearance was consistent with FDS of live adult filarial worms [7, 8].

MRI of the face and neck was subsequently performed on a Philips Achieva 1.5 Tesla MR scanner (Philips Healthcare) using a surface coil (SENSE Flex-S dual element coil, Philips Healthcare).

This study was undertaken to identify any other clinically occult sites of involvement in the head and neck and to evaluate the cervical lymph nodes. MRI (Figure 3) revealed a well-defined lesion over the right zygomatic arch region that appeared isointense on T₁

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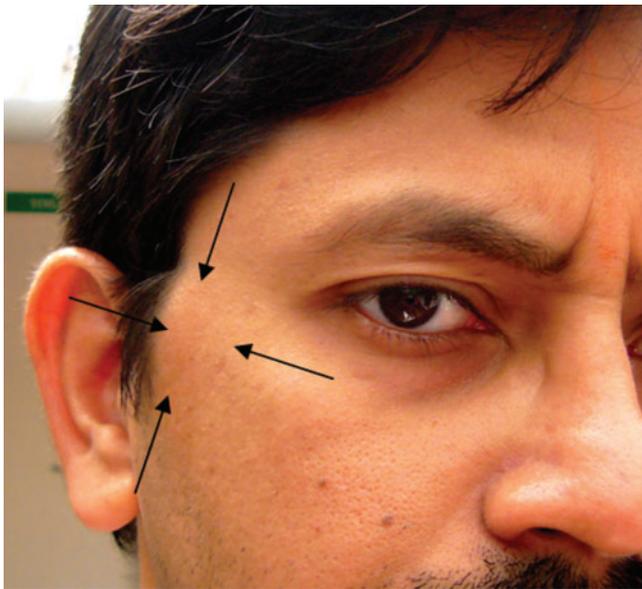


Figure 1. Small smooth swelling over right zygomatic arch (arrows).

weighted images (repetition time (TR): 423 ms, echo time (TE): 16 ms) and hyperintense on short tau inversion recovery (STIR) images (TR: 3480 ms, TE: 23 ms), confirming the cystic nature of the lesion. There was no evidence of cervical lymphadenopathy noted.

Based on the imaging appearances, a diagnosis of live filarial infection in facial subcutaneous soft tissues was suggested. A nocturnal peripheral blood smear showed evidence of mild eosinophilia (16%) without any presence of microfilariae. Urine examination and microscopy was negative for chyluria, and urine concentrate did not show presence of microfilariae. Tests for filarial antibodies and antigens were not available at the time in our institution and hence were not carried out.

Operative and pathological findings

During surgery a well-defined cystic lesion with thick capsule was excised from the soft tissues overlying the right zygomatic arch. It revealed straw coloured fluid

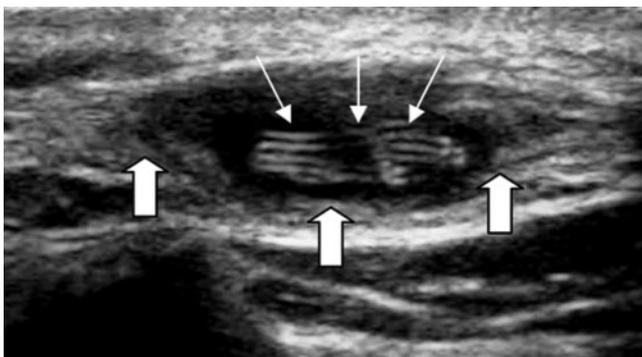


Figure 2. Greyscale sonogram showing a thick-walled, hypoechoic, subcutaneous cystic lesion (large arrows) with multiple linear echogenic structures (small arrows). These linear structures showed persistent twirling motion on real-time imaging.

and multiple whitish thin tubular worms within (Figure 4a), some of which were still moving after the lesion was incised. The length of the adult filarial worms ranged from 45 to 63 mms.

Photomicrogram showed intense inflammatory reaction with eosinophils, few lymphocytes, granular debris and embedded adult filarial worms (Figure 4b).

Discussion

Lymphatic filariasis is transmitted by the *Culex* mosquito and caused by two closely related nematodes, *Wuchereria bancrofti* and *Brugia malayi*, which are responsible for 90% and 10% of infected cases, respectively [3, 4]. Adult worms are found in the lymphatic vessels and lymph nodes only in humans; there is no animal reservoir [2, 3]. These adult worms live in the lymphatic vessels of the definitive host and microfilaria are released and circulated in the peripheral blood. Most frequently involved lymphatics are those of lower limbs, retroperitoneal tissue, spermatic cord, epididymis and mammary glands [5, 6].

Filariasis causes a spectrum of diseases including asymptomatic microfilaremia, acute lymphangitis and lymphadenitis, chronic lymphadenitis, oedema of limbs and genitalia and tropical pulmonary eosinophilia [9]. Diagnosis of the disorder is based on the appearance of microfilariae in blood smears and occasionally in hydrocoele fluid or chylous urine [10].

The FDS on ultrasound was first described in 1994 by Amaral et al [7]. They described the movements of live adult filarial worms in the lymphatic vessels as "peculiar, random-appearing movements of objects inside a vessel-like structure". On ultrasound the worms are seen as linear echogenic structures with persistent, random, almost tireless twirling movements.

There have been reports of single or selected number of cases of microfilaremia at various sites such as lymphnodes, breast lumps, thyroid gland, bone marrow, bronchial aspirate, nipple secretions, pleural and pericardial fluid, ovarian cyst fluid and cervico-vaginal smears [11, 12]. Chaubal et al described the FDS as a reliable indicator of filarial infection in the scrotum [13].

Review of English language literature revealed a case of microfilariae causing lymphadenovariex in the head and neck region. This was a 10-year-old Indian male who presented with an asymptomatic cystic neck mass of 8 months duration. Aspiration of the swelling demonstrated numerous *W. bancrofti* microfilariae [14]. Schick et al [15] also reported a case of a cystic lymph node mass in the neck that was verified histologically to be a cystic manifestation of filariasis.

Our case appears to be the first extralymphatic manifestation of live filarial infestation in the head and neck region. There was no associated cervical lymphadenopathy seen on imaging.

Microfilariae appear in the tissue fluid and exfoliated surface material owing to lymphatic and vascular obstruction [16, 17], and their subsequent extravasation may explain the presence of microfilariae in pericardial fluid, breast cyst fluid and bronchial aspirate. Some authors have suggested an alternative explanation for the presence of microfilariae in thyroid aspirate as the

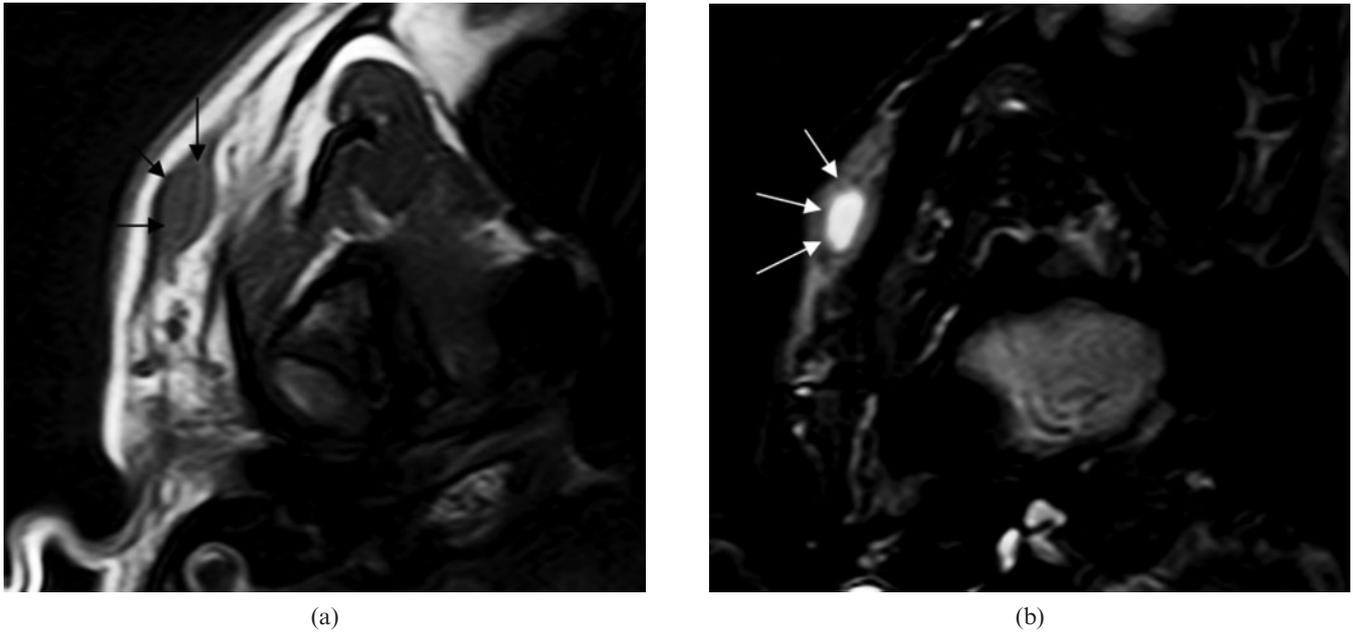


Figure 3. (a) Axial T_1 weighted and (b) short tau inversion recovery (STIR) MRI show a small cystic, encapsulated lesion in the subcutaneous soft tissues overlying the right zygomatic arch (arrows).

result of the parasite lodging within the thyroid microvasculature, which subsequently ruptures [12, 18].

Adult male filarial worms of *W. bancrofti* measure 25–40 mm in length and adult females measure 80–100 mm in length, thickness ranges from 0.1–0.3 mm. Microfilaria measure 200–300 μm in length [19]. Microfilariae are theoretically beyond the resolution of high-frequency ultrasound transducers.

Our patient appears to be an asymptomatic carrier with larvae present in microvasculature. A rupture of vessels may 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.

Conclusion

The FDS seen on high-resolution sonography is diagnostic of live filarial infection. In an endemic region, identification of the FDS in any normal anatomical

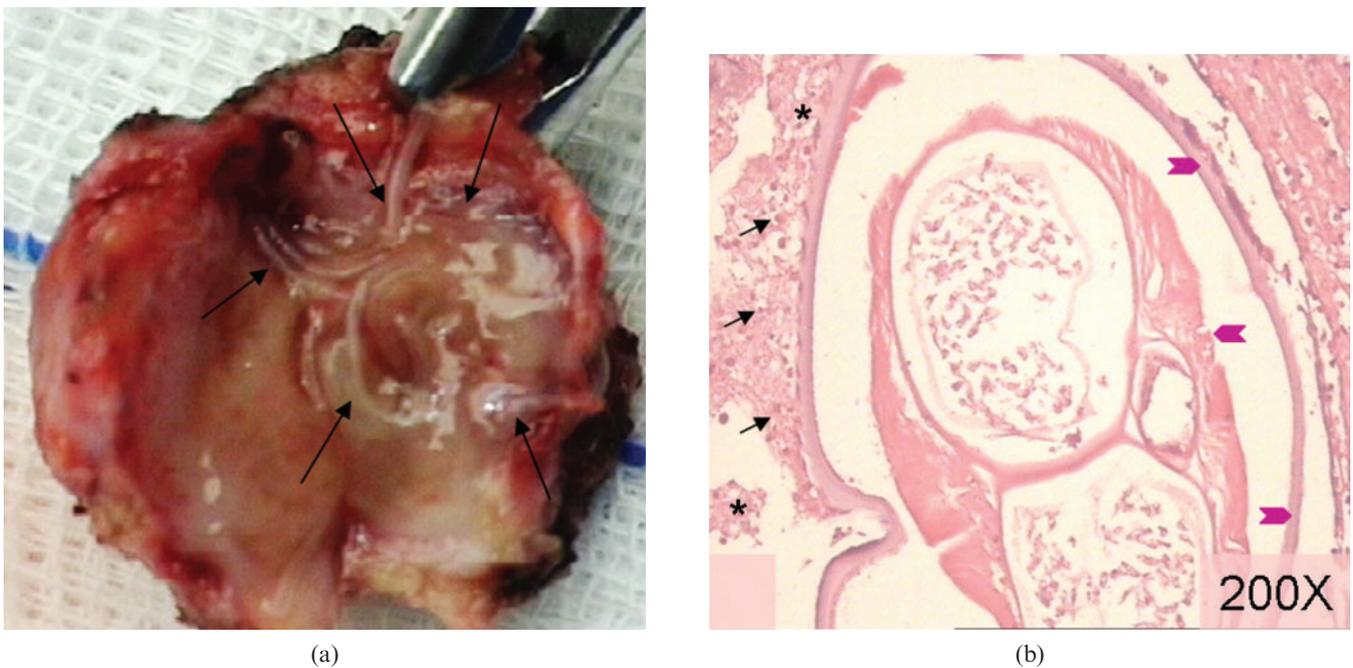


Figure 4. (a) Immediate post-operative specimen showing multiple whitish, tortuous, linear adult worm-like structures (arrows) within the cystic lesion. (b) Photomicrogram shows intense inflammatory reaction with eosinophils, few lymphocytes (thin black arrows), granular debris (black asterisk) and embedded adult filarial worm (arrow heads).

structure or abnormal swelling; however, remote or unusual in location within the body, should strongly suggest the diagnosis of live active filarial infestation. In view of the increasing migratory trends in the global population, it is imperative for radiologists in all countries to be aware of the typical imaging findings of this disease in order to arrive at the right diagnosis.

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