

Armillifer armillatus infection

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Abstract

We report a case of human pentastomiasis in a 70-year-old retired long-distance driver/farmer whose diagnosis was made incidentally while being investigated for a 1-month history of cough and abdominal pain. The chest X-ray revealed multiple comma-shaped and rounded opacities in keeping with Armilliferinfection, most likely *Armillifer armillatus*. The patient made an uneventful recovery after a 10-day course of mebendazole (an anthelmintic) tablet and ciprofloxacin (antibiotic) capsules and was discharged home. He is presently being followed up. This is the first case we have seen in our medical unit thus we are reporting it.

Key words: Armillifer armillatus, pentastomiasis, Nigeria

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Introduction

Armillifer armillatus (*A. armillatus*) infection is an unusual parasitic zoonotic infection transmitted to humans through ingestion of the ova of *A. armillatus* from improperly cooked meat or fresh blood of different species of Large snakes or consumption of foods contaminated by the respiratory secretions of this final host.

As this infection is a rarity in the south- Eastern part of Nigeria where the consumption of snake meat is a taboo, we decided to report this case diagnosed radiologically from our centre.

Case Report

A 70-year-old retired long-distance driver/farmer presented to our unit in July 2009 with a 1-month history of cough, abdominal pain and low-grade fever. Cough was productive of yellowish, mucoid but noncopious sputum. It was associated with nonpleuritic bilateral lower chest pain. However, there was no breathlessness, hemoptysis, drenching night sweats and significant weight loss.

The abdominal pain started insidiously, being localized

at the right hypochondrial area. It was not radiating, not aggravated by coughing and not associated with any other significant gastrointestinal, cardiovascular, genitourinary or neurological symptoms. The past medical and surgical history was not significant. He used to be a long-distance driver and later on a self-subsistent farmer for 10 and 16 years, respectively. He denied any history of ingestion of snake meat either as bush meat or otherwise and in places where he lived noted minimal snake activity.

On examination, he was found to be generally stable, afebrile to touch, not pale, anicteric, no peripheral edema or lymphadenopathy with normal findings in the respiratory, cardiovascular and neurological systems. Abdominal examination revealed nontender hepatomegaly, 6cm below the right costal margin. It was firm, smooth surfaced with a liver span of 14 cm. Spleen was also enlarged 4cm below the left costal margin while the kidneys were not ballotable and there was no demonstrable ascites.

Investigation results showed a packed cell volume of 0.34, total white blood cell count of $3.9 \times 10^9/l$ with

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differential neutrophils (N=70), lymphocytes (L=20%), eosinophils (E=2%, Sputum microscopy, culture and sensitivity yielded no bacterial growth after 24 hour culture and Zeihl Nielsen (ZN) staining for acid-fast bacillus (AFB) was negative in three sputum samples. Liver function tests, renal function, urinalysis and calcium levels were all normal. Chest X-ray showed multiple comma-shaped and rounded opacities with radiolucent center scattered throughout the lung fields in keeping with parasitic infection of *Armillifer* species most likely *Armillifer armillatus* [Figure 1], plain abdominal radiograph also showed calcific lesions [Figure 2]. Abdominal ultrasound scan showed enlarged liver with multiple irregular to-roundish echogenic shadows scattered on both right and left lobes of the liver. The other intraabdominal organs were sonographically normal.

A radiological diagnosis of *Armillifer* infection most likely with *A.armillatus* was made and the patient was given a 10-day course of ciprofloxacin tablets 500 mg twice daily,



Figure 1: CXR of the patient showing multiple comma-shaped calcifications in both lung fields



Figure 2: Plain abdominal radiograph of the same patient

mebendazole 100 mg twice daily and hematinics. He got better with resolution of his presenting symptoms and was discharged home. He has been followed up in the medical outpatients and is presently stable.

Discussion

A. armillatus is a pathogen causing human pentastomiasis. This is an uncommon parasitic zoonosis and to the best of our knowledge, this case is the first being reported from the South Eastern part of Nigeria. Human infection is caused by the larval stages (nymphs) of the arthropod-related tongue worms *Linguatula serrata*, *A. armillatus*, *A. moniliformis*, *A. grandis*, *Porocephalus crotali*. Worldwide, the pentastomid genus *armillifer* is the most common in human pentastomiasis. There are four pathogenic species of *armillifer*: *A. agkistrodontis* in China, *A. armillatus* in Africa and the Arabian Peninsula, *A. grandis* in Africa and *A. moniliformis* in Southeast Asia.^[1] The majority of cases have been reported from Africa, Malaysia and the Middle East where diagnosis was mainly made on autopsy and less often in China and Latin America.^[2] In Europe and North America the disease is only rarely encountered in immigrants and longterm travelers and the parasitic lesions may be confused with malignancies.^[2]

This infection was first reported in Nigeria in 1975 by (Azinge *et al*,^[3] in Benin-city). The patient was being investigated for miliary tuberculosis and his chest radiograph showed the classical comma shaped calcifications due to *A. armillatus* infection. The focus of his infection was studied by contact tracing of the household and of the subject. Ten members of the family were screened radiologically and 40% were found to have calcifications due to *A. armillatus*. Another report from Ilorin (South-west Nigeria), showed *A. armillatus* calcification in three cases (1.4 %) out of 214 plain abdominal radiographs analyzed in a tertiary hospital there.^[4]

Armillifer pentastomids propagate cyclically between snakes to mice and snakes to other mammals but snake is the main source of infection of human pentastomiasis. Infection depends on the scope of snake's activity and human's dietary habits. It can be acquired through drinking fresh blood of serpent with contaminated eggs, eating raw or undercooked serpent flesh, eating egg contaminated vegetable or drinking pond water contaminated by infected snakes and finally the adult pentastomids may be ingested by man, while mammals can eat adults expectorated by frightened snakes. In our patient, there was no history of snake meat consumption; however, it is possible that he could have been infected through the other routes listed above. In the first reported case in Nigeria there was also no history of snake meat consumption in all the family members who had the calcified nymphs in their chest radiographs. The route of

their infection was not elucidated clearly but we know that snakes are endemic in that part of the country and their infection may be through the other routes listed above.

In the GIT of human host, the ova hatches into minute 4 legged primary larva and invades the viscera. After encapsulation by host tissue for several months, infective larval stage develops. Nymphs finally resemble adult legless vermiform pentastomes in shape. This infective larva causes the pathological hallmark of *A. armillatus* infection. They invade target organs via migration from small gut through peritoneum. Pleural infection could be through the diaphragm and calcification has not been described in skeletal muscles. They have been known to cause marked dystrophic calcifications in affected organs and further organ changes depend on whether larva is alive or dead.

In human patients, three types of pentastomid lesions have been described; this was first described by Ma et al,^[5] and later by Tappe and Büttner^[2]

- Viable nymph of *A. armillatus* with thin fibrous capsule encysted in lung with little or no cellular infiltration or antigenic compound elaboration.
- Necrotic pentastomid granuloma. Here a dead nymph is found with large amounts of antigen released from the dying nymph and activation of many immune cells. The central necrosis is surrounded by a thin wall of hyalinized epitheloid cells, with large area of fibrous tissue and a few giant cells, macrophages, lymphocytes, plasma cells and often many eosinophils. This is the most common type of lesion in longstanding infections.
- Granulomatous scar or cuticle granuloma without typical structures of pentastomes and little or no antigens are released. Cshaped fibrotic lesion, lymphocytes, but no eosinophils are found.

Most human infections are asymptomatic and disease may be an incidental finding during routine medical consultation or at autopsy. For the patient presented it was an incidental finding during a diagnostic workup for his chronic cough.

The symptoms a patient presents with depend on the organs involved. Patients may have chronic cough, abdominal pains as in our case or intestinal obstruction requiring surgical intervention.^[6] In heavy infections with *Armillifer* spp., death may occur due to secondary septicemia, pneumonia or severe enterocolitis.^[5,7]

Diagnosis is mainly based on a characteristic radiograph or histopathological examination of biopsied lesions. The radiological appearance of calcified nymphs of *A. armillatus* is among the most characteristic of all soft tissue calcifications. Typical multiple commashaped calcifications

on the chest or plain abdominal radiograph are seen. This, however, could be confused with calcifications due to cysticerci but the latter is restricted to the skeletal muscles and brain and never occurs within the thoracic or abdominal cavity^[8] Ancillary investigations include full blood count which may show mild eosinophilia and ESR may be high. Few serological studies have been conducted; an ELISA and Western blot have been employed to confirm the presence of *A. armillatus* in a snake farmer;^[9,10] however, no serological test is readily available. Recently PCR tests have been used for diagnosis of pentastomiasis from a clinical sample but this too is not readily available.^[9] Thus, visceral pentastomiasis often remains a histopathological diagnosis and correct diagnosis is necessary in order to rule out malignancy and to treat patients with trematodal, cestodal or nematodal infections accordingly.^[2] In asymptomatic patients no treatment is necessary, since parasites degenerate after approximately 2 years. Only in symptomatic infections with numerous parasites may a surgical approach be considered. No established antiparasitic chemotherapy is available for pentastomiasis; however, Mebendazole has been suggested for those with mild to moderate infestation. It is not clear whether it was the antihelminthic or the antibiotic given to our patient that helped him but he made a marked improvement with the resolution of the cough and abdominal pain. The patient, however, is being followed up closely in our outpatient unit.

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