Case Report: Zoonotic Anatrivosomiasis in an Illinois Resident

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Abstract. We describe a case of zoonotic anatrivosomiasis in a patient from Illinois. A 44-year-old immigrant from Mexico originally presented with a history of multiple oral ulcers and two submucosal nodules on the dorsal surface of the tongue. An incisional biopsy was taken to assist with diagnosis. Examination of stained sections revealed the presence of a coiled nematode. The histologic examination displayed trichuroid features. Anatomic structures that aided in the identification included esophagus embedded in a prominent stichosome in the anterior end, paired bacillary bands, and small size. The location of the worm within the oral mucosal epithelium also facilitated the diagnosis.

INTRODUCTION

Anatrivosoma species are small trichuroid nematodes that are an unusual and poorly known group of parasites.¹ Several species have been described, including *A. buccalis* from the mouth of opossums (*Didelphis*) in the United States, *A. cutaneum* of the skin and nasal mucosa of primates in Africa and Asia, *A. cynomolgi* of the nasal mucosa of rhesus monkeys in Southeast Asia (this species has been reported from a number of different macaque monkeys in Southeast Asia), and *A. ocularis* from the orbit of tree shrews in Southeast Asia. The life cycle is unknown, although embryonated eggs are sloughed in trails, presumably swallowed and passed out of infected animals in the feces. If the life cycle is direct or involves an arthropod intermediate host has been studied but never elucidated.

The anatrivosomes share many features in common with the other trichuroids, including coelomyarian/polymyarian musculature, prominent stichosome composed of individual stichocytes in which the esophagus is embedded, and paired bacillary bands in the lateral fields.¹ They tend to be relatively small slender worms, and the reproductive system in both males and females is a single tube. On occasion, at least in *A. buccalis*, portions of male worm have been seen in the uterine canal of female worms in tissue sections.

We report our findings on a patient in whom we identified zoonotic *Anatrivosoma* infection. This is apparently only the fourth such case and the first that we are aware of describing the parasite in the mouth (the other three report cases from the skin).

CASE REPORT

The patient is a 44-year-old Hispanic man who has lived in Illinois for 20 years. He grew up in Guanajuato, Mexico, which is in central Mexico about 150 miles north of Mexico City. He moved from Guanajuato about age 21 and then, worked in the California farm industry for 2 years. At that point, he moved to Illinois where he still resides. Recent travels include: Piedras Negra, Coahuila, Mexico in both July 2008 as well as July 2009. (Piedras Negra is just across the Rio Grande river from Eagle Pass, Texas); he also visited Guanajuato in January 2009. The

patient currently works as a landscaper, although he has also worked in a foundry.

Medical history reported that he had been diagnosed with hypertension in the past but was not taking any systemic medications. He denied any medicinal or environmental allergies. Patient had initially seen his primary-care physician for symptoms. He had initially presented with a chief complaint of the feeling of a lump in his throat, although lesions on his tongue and lips began to appear early in the disease course. Lesions were reported to be relatively transient, lasting 4–5 days. The primary-care provider referred the patient to an otolaryngic specialist for further evaluation of throat symptoms. The otolaryngologist subsequently referred the patient to an oral pathology specialist with a differential diagnosis including cicatricial pemphigoid and viral disease. Various medications had been tried without benefit for the chief complaint, including esomeprazole for possible gastroesophageal regurgitation, topical fluocinonide and systemic prednisone for possible autoimmune ulcers, various antibiotics (azithromycin, amoxicillin, and amoxicillin with clavulonate), and nystatin for possible mycotic overgrowth.

At clinical presentation, the patient had what appeared to be multiple oral and lip ulcers, which appeared to be in the process of healing (Figure 1), and two indurated submucosal nodules on the dorsal surface of the tongue that each measured approximately 4 mm in diameter. There were some possible tracks on the upper and lower lip lesions (Figure 2). At time of biopsy, the clinical impression was that of chronic granulomatous disease versus amyloidosis. An incisional biopsy of the tongue was performed in an area that was somewhat indurated, and the patient said that he felt it was the newest area and felt that it would soon ulcerate.

By time of biopsy, symptoms had been present for about 3 months. A tissue sample from the tongue biopsy was sent for routine histologic sectioning. Review of hematoxylin and eosin (H&E)-stained sections revealed the presence of a coiled female nematode lying under and within the epithelial mucosa (Figure 3).

A more detailed study of the worm provided the following morphologic features that were compatible with a diagnosis of *Anatrivosoma* spp. The maximum diameter, measured in the region of the reproductive tube, was no more than 70 μ m; stichocytes were prominently visible in the anterior end, even at lower magnification, and they contained the small tubular esophagus, the presence of two bacillary bands and compatible morphology of the muscle cells, and the reproductive tube (Figures 4 and 5).

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FIGURE 1. One of several ulcerated lesions on the lip, some of which were in various stages of healing. This figure appears in color at www.ajtmh.org.

Although only a single worm was evident in the sections available for study, the impression at the time of examination was that multiple worms were likely present.

The patient was treated with 100 mg of mebendazole two times daily for 20 days, and a positive therapeutic outcome was achieved. All oral lesions resolved with no recurrence of symptoms with 1 month follow-up. At one point, the patient described skin lesions during mebendazole therapy, although they were thought by the patient's primary-care physician to be unrelated to the current discussion.

DISCUSSION

Although sharing many features with other more common trichuroid nematodes, very little is known about the biology, life cycle, host range, and degree of speciation of *Anatrichosoma*. The adult worms live in superficial tunnels of the epithelium of the skin, nasal mucosa, or mouth, where the female worm lays fully embryonated eggs that are barrel-shaped and have the typical bipolar plugs common to other trichuroids. Eggs are sloughed directly into the environment or swallowed and passed in feces. Attempts to establish experimental infections by orally dosing animals or by exposing eggs to common

FIGURE 2. Track-like lesions seen on the inner cheek. This figure appears in color at www.ajtmh.org.

FIGURE 3. The coiled, non-gravid female worm is clearly visible in the sections of the tongue biopsy. At this magnification, some features of the worm, notably the stichocytes, are evident. Scale bar = 200 μ m. This figure appears in color at www.ajtmh.org.

insects such as beetles and cockroaches have uniformly been unsuccessful; thus, the mode of infection to the next host is unclear (Orihel T and others, unpublished data).

Only a handful of human cases of infection with anatrachosomes have been reported in the literature—two cases from Japan,² one case from Vietnam,³ and one case each in Malaysia⁴ and Italy.⁵ Four of the previous cases occurred as skin lesions, whereas in the case from Italy, the parasite was noted incidentally in a breast biopsy containing *Dirofilaria repens*. This report is the first involving the mouth. This suggests, but does not confirm, that the present case may be caused by *A. buccalis* of the opossum. Given the distribution of that species, the infection could have been acquired in Mexico, although the recent onset of symptoms argues that it was acquired locally in Illinois. However, there are likely other species of

FIGURE 4. Distinguishing morphologic features are clearly evident in this section and include bacillary bands in the lateral fields on both sides of worm (arrow to band on right side) and the small esophagus (arrowhead) embedded in the stichocyte (asterisk). The nature of the muscle cells and cuticle are also visible. Scale bar = 25 μ m. This figure appears in color at www.ajtmh.org.

FIGURE 5. In these sections, posterior to the stichosome/esophageal region, the single female reproductive tube is visible, including infertile, undifferentiated eggs (arrows). Scale bar = 25 μ m. This figure appears in color at www.ajtmh.org.

Anatrachosoma that have not been recognized and described, and therefore, for the present time, it may be most appropriate to leave this as *Anatrachosoma* sp. This is especially true in light of two cases of unusual dermatitis in a dog and a cat in the United States caused by *Anatrachosoma*.^{6,7}

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