

Case Report of and Description of Parasite in *Mammomonogamus laryngeus* (Human Syngamosis) Infection

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About 100 cases of human infections with *Mammomonogamus laryngeus* (Syngamidae, Syngaminae) have been reported, with virtually all cases originating in the Caribbean Islands and Brazil. This report describes the fifth patient in North America infected with *M. laryngeus* and the first case documented to originate in Jamaica. The patient complained of a characteristic persistent nonproductive cough and a lump in her throat. *M. laryngeus* is a nematode in which the male and female are permanently joined in copula, producing a distinctive Y shape. Since there is scant clinical information about this parasite, this report includes a description of the adult worms and eggs, a summary of the epidemiology, and the clinical manifestations in humans. Illustrations are presented to facilitate future identification of this parasite and to expedite treatment.

Mammomonogamus infection in humans is quite rare, with only about 100 cases reported to date. Most cases have originated in the Caribbean Islands or Brazil, and about half of these cases have originated in Martinique (14–16). All but two cases thus far reported have occurred in residents of the Caribbean area or in visitors to that region. The two exceptions were one patient from the Philippines and one from Thailand (15, 19). Patients diagnosed with syngamosis in France, England, and Australia all acquired the parasite in the Caribbean area (4, 5, 7, 9, 17).

Only five North American patients have been diagnosed with *Mammomonogamus laryngeus* infection, and all of them also visited the Caribbean area shortly before the onset of clinical symptoms (8, 11, 20, 21). The case described here, which occurred in Ithaca, N.Y., is the fifth case to be reported in North America and the first case of infection as far as we know to have been acquired in Jamaica (9).

The clinical symptoms are frequently characteristic. Patients often complain of a chronic nonproductive cough, asthma-like symptoms, or a crawling sensation in their throats (4, 5, 9, 11, 18, 20, 21).

Since so few cases of human infection have been reported, most microbiologists and physicians are unaware of this parasite. Because there is very little information in infectious-disease reference books on this infection, the purpose of this report is to increase awareness of this parasitic illness, to provide illustrations to assist in the identification of these worms, and to report the fifth case of *M. laryngeus* infection in North America. In addition, we extend the geographic range of the organism to include Jamaica.

Case history. A 25-year-old female presented to an outpatient clinic with a headache, a sore throat, and a dry cough 8 days after returning from a 1-week trip to Jamaica. While in Jamaica, she stayed in a tourist hotel. Although she ate only at the resort, she did have fresh salads there. She had no known exposure to live ruminants. She was initially treated by a clinic physician for viral bronchitis with a cough suppressant. Subsequently, she saw her private physician, who prescribed a 10-day

course of oral erythromycin. Although her major symptoms resolved, she continued to have paroxysms of nonproductive coughing and low-grade fevers of 99 to 100°F (ca. 37.2 to 37.8°C) over the next 4 months.

After she complained of a persistent lump in her throat, her husband observed a worm-like object in the back of her throat. She returned to the clinic, where a red-brown Y-shaped worm-like structure was removed by forceps from the left posterior pharynx. At this time she had a normal chemistry profile, urinalysis, and complete blood count, with only a single eosinophil observed in the differential leukocyte count.

Her symptoms initially resolved after removal of the worms; however, a few days later, intermittent dry coughing recurred. A stool sample for ova and parasites was negative. Because coughing persisted, a bronchoscopy was performed 4 months later. No adult worms were seen and no ova were found in concentrate preparations made from bronchial washings. She has had no further complaints since her bronchoscopy, and no further treatment has been instituted.

Description of parasite. Identification of this worm is most easily made grossly by the typical Y-shaped appearance of the nematodes in copula. Species-level identification is based on the size of the worms, the appearance of the mouth parts, the shape and length of the female's tail, the presence or absence of spicules in the male, and the size and appearance of the eggs (12). The gross specimen can be preserved in 70% ethanol, 70% isopropyl alcohol, or 10% formalin. It can be cleared for optical microscopy by using a phenol-alcohol solution (80 ml of liquified phenol–20 ml of 100% ethanol).

This worm is blood red to reddish-brown because of hemophagia. The Y shape consists of two nematodes, with the female constituting the long arm and base of the Y. The shorter arm is the male worm, whose bursa is attached to the side of the female worm at the level of the vulva. Both males and females have a mouth, or buccal capsule, which is thick-walled, contains short or long ribs, and has eight small teeth at its base. There is a cuticular rim around the oral opening, surrounded by lips. The male worms are 3.0 to 6.3 mm long and 360 to 380 μm wide with or without spicules. Spicules are 23 to 30 μm long. Female worms are 8.7 to 23.5 mm long and 550 to 570 μm wide. The vulva is in the anterior part of the body. The tail may be long or short. Eggs are ovoid to ellipsoid,

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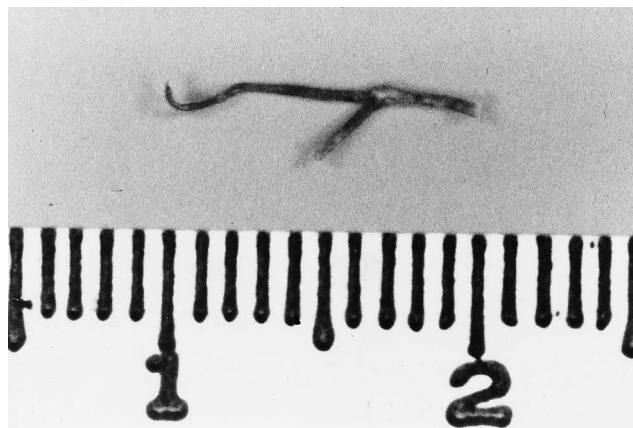


FIG. 1. The gross appearance of the fresh specimen is the characteristic Y form created by the permanent copulation of the smaller male worm and the larger female. The fresh worms are red-tan because of hemophagia. The scale is in centimeters.

are nonoperculated, and measure 78 to 95 μm by 42 to 54 μm (12).

The worms recovered in this case were in copula (Fig. 1). The male worm was 4.3 mm long with a maximum width of 0.43 mm. The female worm was 14.0 mm long with a maximum width of 0.55 mm. A thick muscular wall, the cuticular rim, and several teeth were seen in the buccal cavity (Fig. 2). Eggs in the female measured 75 to 80 μm by 45 to 60 μm (Fig. 3).

Since the sexes are joined in permanent copulation, nematodes of the family Syngamidae present a very distinctive gross Y-shape appearance. Worms of this family are found in birds (genera *Syngamus* and *Cyathostoma*) and in mammals (genera *Mammomonogamus* and *Rodentogamus*), especially in tropical and subtropical regions. The genus *Mammomonogamus* is a common parasite of domestic ruminants, particularly cattle, and of cats (3).

M. laryngeus (synonyms, *Syngamus laryngeus* and *Syngamus kingi*) from cattle and other ruminants is the species reported to be involved in human infections. This species varies in appearance and at times has been considered two species: *M. laryngeus* and *Mammomonogamus nasicola*. At present, both forms are considered to be the same species, *M. laryngeus* (13).

Although it is in the same order (Strongylata) as hook-

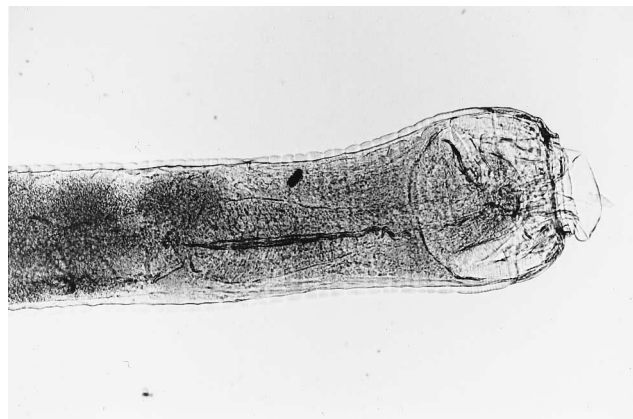


FIG. 2. Unstained phenol-cleared specimen of the female worm. The head contains the mouth parts. The esophagus is also well illustrated. Magnification, $\times 16$.

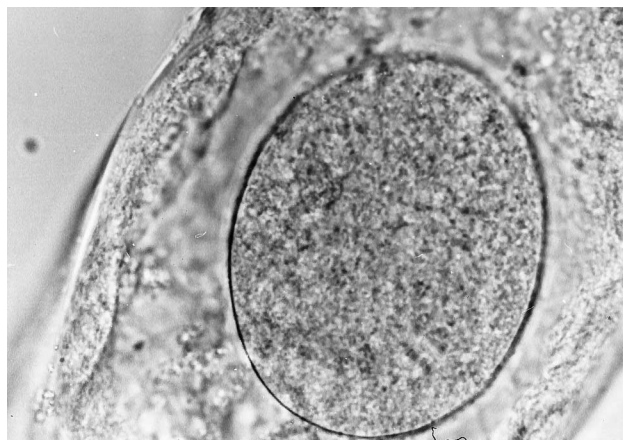


FIG. 3. Unstained phenol-cleared specimen of a mature ovum. The ovum has a thick cortical shell and is nonoperculated. Ova measured 75 to 80 by 45 to 60 μm . Magnification, $\times 400$.

worms, *M. laryngeus* is in a different superfamily (Strongyloidea) because it lacks the teeth and cutting plates that are located at the orifice of the buccal cavity in hookworms (superfamily Ancylostomatoidea). In *M. laryngeus* the teeth are small, are located deep in the buccal cavity, and are not used for attachment to the host (2).

Epidemiology. Because *M. laryngeus* is a common veterinary parasite, its identification can be confirmed by an experienced veterinary parasitologist or pathologist. Other resources for identification include other professional parasitologists, the Infectious Disease Branch of the Armed Forces Institute of Pathology, and the Communicable Disease Center in Atlanta, Ga.

The life cycle of *M. laryngeus* has not been fully clarified, but it may be similar to that of the bird parasite *Syngamus trachea*. The adult worms are normally found in the laryngotracheal region of ruminants. The eggs are coughed up and swallowed, so they may be recovered in sputum or more often in feces. Outside the host, the eggs undergo development until they contain an infective larva. This larva may stay in the egg or be released into the environment. Ruminants become infected by ingesting food or water containing the infective larvae. Ingestion of carrier hosts such as earthworms, snails, or arthropods may be another source of infection. In the digestive tract, it is assumed that the larvae are released, cross the intestinal wall to mesenteric veins, and migrate to the tracheolaryngeal area, where the adult worms mate and start reproducing (1).

Clinical manifestations, diagnosis, and treatment. In humans, unlike in ruminants, infection with *M. laryngeus* is usually associated with a severe chronic nonproductive cough. This may be associated with a crawling sensation and hemoptysis (11, 15, 21). Some patients have had severe asthma symptoms, probably due to airway obstruction by organisms in bronchi or bronchioles (4, 5, 9, 18, 20). Two patients were reported to have transient pneumonitis, suggesting a possible pulmonary phase (18). No other evidence for systemic infection has been reported. In most patients, only a single pair of worms is found. However, in eight patients, multiple worms were found (14, 15), including one patient from Thailand from whom 96 pairs of worms were recovered in 4 days (15).

The symptoms of the patient described here began 8 days after she returned from Jamaica. This is consistent with a reported incubation period of 6 to 11 days (21). Because the time for development of an *S. trachea* larva into an adult worm

in avians is said to be 3 weeks (21), it is likely that our patient was infected by adult worms.

Patients with *M. laryngeus* infection have few abnormal laboratory test results. Eosinophilia has been an inconsistent finding. Among 12 patients for whom eosinophil data were provided (4–6, 10, 11, 15, 18, 20, 21), only four patients had eosinophil counts greater than 6%, or eosinophilia (6, 10, 18). There was no correlation between the number of worms recovered and eosinophilia. In fact, the patient with the most worms had only a 3% eosinophil count (15). The relatively low eosinophil counts probably reflect the lack of host tissue invasion by the parasite, since the adult worms live on the epithelial surface.

Infection can be detected when the adult worms are coughed up, seen on direct examination of the throat, or visualized by endoscopic techniques. Eggs may be recovered in the sputum and/or in the stool. In one patient, diagnosis was made by examining histologic sections of an endobronchial cyst containing "a 1-inch . . . string" removed by bronchoscopy (8).

Treatment of *M. laryngeus* infection has varied. One patient was cured by simply coughing up a pair of worms (21). In some patients, cure was effected by manual or bronchoscopic removal of one or more worms (5, 8, 14, 18). Removal of the adult worm(s) generally resulted in resolution of symptoms. However, some patients have also received a course of anthelmintic therapy (4, 6, 9–11, 14, 16, 20), but there has been no controlled study of this therapy. The drugs most commonly used have been thiabendazole and mebendazole. Doses have ranged from 200 to 3,000 mg/day for as little as 3 days and as much as 20 days. In all cases, when patients were treated with anthelmintic drugs, the patients were described as cured.

Summary. *M. laryngeus* infections in humans are unusual and almost always associated with residency in or visits to the Caribbean Islands or Brazil. However, considering the number of North Americans who visit the Caribbean, one would anticipate seeing more examples of this infection. The description and illustrations of *M. laryngeus* presented here should facilitate proper identification of the parasite and expedite appropriate clinical management of the patient.

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