SHORT REPORT: A CASE OF ECTOPIC SCHISTOSOMIASIS IN PUERTO Rico WITH SOME OBSERVATIONS ON THE BIOLOGY OF THE PARASITE

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Abstract. A recently acquired Schistosoma mansoni infection that resulted in a cervical polyp containing a pair of adult worms is reported in a Puerto Rican woman. Active schistosome transmission is not commonly reported in Puerto Rico at the present time and the ectopic location of the worms is rare in very light infections. Observations on the biology of the parasite recovered from the patient are described.

Active Schistosoma mansoni infections, which once had a significant presence in Puerto Rico, are no longer common. However, schistosomiasis is not a reportable disease and therefore it is not possible to estimate current prevalence or rates of transmission. Because the adult worms can live for 20 years or longer, infections of indeterminate age are the usual finding. The present report deals with a case that we believe was acquired within a year of its detection and that resulted in ectopic schistosomiasis of the uterine cervix, a rare finding in Puerto Rico even when the parasite was common.1

A 43-year-old woman came to a gynecologic clinic complaining of an itchy vaginal irritation and intermittent vaginal bleeding of fairly recent onset. The patient had a history of nonspecific endometritis. An endocervical polyp was observed and excised by the gynecologist. Histologic examination of multiple sections of the excised tissue revealed a pair of adult schistosomes morphologically consistent with S. mansoni in a vein and viable eggs in the adjacent tissue with cellular inflammation, eosinophils, but little fibrosis (Figure 1). Four fecal samples collected over a six-week period were examined in duplicate and each consistently revealed seven S. mansoni eggs/g.2 Serologic examination revealed a very weak circumoval precipitin reaction.3 However, multiple reactive bands were detected against S. mansoni worm and egg proteins on Western blots.3 No other symptoms were evident and the results of an ultrasound examination were normal.

Interviews established that the patient was Puerto Rican but that she lived in the continental United States for more than 20 years (New York City) and had recently come to live in the San Juan metropolitan area (two years). She had never traveled to any other area endemic for schistosomiasis. Contact with natural waters was very rare and she could recall each trip to rural areas. Symptoms were recounted that were consistent with S. mansoni infection within the previous year. The only site where water contact and presumed exposure to cercariae occurred was examined. This site was known by the local inhabitants to have been a transmission site some years ago. No Biomphalaria were detected on two visits, although ample snail habitat and opportunity for contamination of the water with human feces were observed as were numerous swimmers. The patient was treated with praziquantel and she returned to the mainland United States.

Because on several other occasions, miracidia derived from local human infections have not been infective or only slightly so for various laboratory strains of B. glabrata designated as Puerto Rican, it was thought to be useful to test the infectivity of miracidia hatching from the feces of this patient for a laboratory strain of snail. Fecal material was collected four times over a six-week period. Miracidia hatched readily and were used to expose M line B. glabrata (NMRI strain obtained from Dr. Fred Lewis, Biomedical Research Institute, Rockville, MD) to five miracidia each. Mice and hamsters were exposed to the resulting cercaria.

Eight of 345 snails exposed and surviving 30 days after exposure shed cercariae but all died within days of the onset of shedding. Infections in rodents were assessed 6–8 weeks after exposure and seven had only female worms; only one hamster developed a bisexual infection. Retrospective analysis determined that only one of eight snails had been shedding male cercariae. Eggs from the tissues of the infected rodent hatched poorly and did not infect any of the 35 snails exposed. Seventy-two and 85% of the M line snails serving as controls for each exposure to a laboratory strain of parasite became infected. Previous attempts to infect laboratory strains of B. glabrata with S. mansoni miracidia obtained from two other human infections in Puerto Rico have yielded similar results.5 This suggests that the adapted parasite strain studied in most laboratories is somewhat different from the parasite currently present in humans.

Instances of recent, active transmission of S. mansoni in Puerto Rico are seldom reported. However, Tsang and others6 have recently reported that many adults are serologically positive for S. mansoni. Numerous changes in prevailing life styles, sewage disposal, water contact, and in the environment have occurred on the island in the last 25 years, most of which are unfavorable for transmission of this parasite. Apparently, transmission is intermittent and or at a low intensity with most infections, as in the present instance, consisting of few worms in otherwise healthy persons and therefore usually go undetected. This case is unusual in that an infection, which apparently consisted of very few worms, resulted in adult worms lodging in an ectopic site, the uterine cervix.

The finding of schistosome eggs in the female reproductive tract, predominately S. haematobium, but also S. mansoni, is not uncommon in Africa,7 and has been reported from Brazil.8 Arean2 reported schistosome involvement of the reproductive tract in Puerto Rican women when he reviewed a large series of 78,238 surgically removed gynecologic specimens and 3,232 autopsy cases from 1925 to 1955 when active transmission of schistosomiasis in Puerto Rico was well documented. Even when heavy infections were common, involvement of the reproductive tract was rare because Arean detected only 18 cases of S. mansoni in the female genital tract. Only six of them involved the uterine cervix. Feldmeier and
Kranz\(^9\) and Feldmeier and others\(^{10}\) have reviewed the world literature on schistosome infections in females. Feldmeier and Kranz cite numerous case reports of schistosomiasis of the female genital tract but they emphasize that “reliable data on the prevalence and morbidity do not exist.”\(^9\) *Schistosoma haematobium* involvement of the female genital tract can be very common in parts of Africa. For example, Renaud and others\(^{11}\) reported a prevalence of 75% vaginal schistosomiasis in Niger. However, the prevalence of *S. mansoni* in any portion of the reproductive tract of females in any of its endemic areas is much less common and has not been as well studied.

Coelho and others\(^6\) found that only four (0.32%) of 1,250 women with diagnosed *S. mansoni* infection had the parasite ova in cervical scrapings. Most diagnoses, as in the present case, are made through incidental observations during gynecologic examinations and schistosome eggs are often associated with benign cervical lesions. The present case is of interest because the infection was apparently acquired in Puerto Rico at a time when transmission was presumed to be interrupted. It is also interesting because both an adult worm pair and eggs were found in a cervical polyp.

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