

## STUDIES ON SCHISTOSOMIASIS MANSONI IN PUERTO RICO \*

### I. THE HISTORY OF SCHISTOSOMIASIS IN PUERTO RICO

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Increasing information on the distribution, pathogenicity and symptomatology of *Schistosoma mansoni* infection in Puerto Rico has indicated more and more the importance of the disease in this Island. Since the discovery by Doctor González Martínez three decades ago of the eggs with lateral spines in the feces and later at autopsy of the parent worms in the inferior mesenteric veins of indigenous cases, a considerable amount of data on this parasite has been added to medical literature. It seems wise, therefore, to review briefly what contributions have been made to the subject by investigators working in Puerto Rico.

In February, 1904, during an investigation on afebrile dysentery, González Martínez studied two young patients in Mayagüez<sup>1a</sup>, who had continuously resided in the area and whom he diagnosed as suffering from intestinal schistosomiasis. The stools of both of these boys contained schistosome eggs with a lateral spine ("huevos de bilharzia con espícula lateral"). This information was communicated to the Medical Association of Puerto Rico on April 3, 1904, and was published as a thirty-two page monograph, copies of which were sent to libraries and prominent investigators. In this publication it was concluded that schistosomiasis was endemic in Puerto Rico and was therefore not confined to the continent of Africa; that it was probably introduced by the importation of slaves from the Congo, Angola and Kafirland; that it was found along the coast of Puerto Rico in association with *Distoma hepatica* which was prevalent in the northern and western parts of the Island; that it was more

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common in males than in females due to the greater opportunity for exposure; that in Puerto Rico, in contradistinction to other regions where schistosomiasis occurred, the intestinal form was more common than the vesical, and that its incidence was less than that of filariasis or hookworm infestation. Six months later (October 11, 1904) González Martínez<sup>1b</sup> had the opportunity to perform an autopsy on one of the patients of the First Anemia Commission of Puerto Rico. In the portal vessels and tributaries 219 schistosomes were found, mostly males but including a few females. The majority of these specimens were sent to investigators abroad, including Doctor John Catto, who had recently discovered a schistosome in the Orient (*S. cattoi*, i. e. *S. japonicum*). In a letter of acknowledgment (December 15, 1904), which the present writer has seen, Catto wrote: "I have shown your worms and papers to Sir Patrick Manson and he is also very interested." Continued search for cases during the year 1904 revealed a total of 59 cases of intestinal schistosomiasis out of 1,321 unselected patients<sup>1</sup>. In January, 1905, another autopsy was performed, this time at Mayagüez, from which four male worms were obtained. These were preserved in formalin and two of them were forwarded to Professor R. Blanchard, with the suggestion that the schistosomes producing enteritis and having lateral-spined eggs discharged in the feces constituted a new species. On July 2, 1905, González Martínez addressed the Medical Association of Puerto Rico on the subject of schistosomiasis, at which time he was able to add Santo Domingo to the endemic area of the Western Hemisphere and estimated the infestation to be as high as 7.8 per cent in the country population of Puerto Rico. It was stated that in all of these cases only the intestinal type with lateral-spined eggs was encountered. Likewise, blood studies by this investigator had shown that the disease produced a mild leukocytosis, moderate eosinophilia (8 per cent) and moderate mononucleosis (10 per cent), but no tendency to marked anemia. In the light of this documented evidence

<sup>1</sup>The First Anemia Commission (Dec. 1, 1904 publication) also found 21 cases of bilharziosis out of a series of 1,408. They stated (p. 91): "Bilharziosis recti is particularly common and is frequently unaccompanied by blood in the feces. There is often, however, a large amount of mucus seen in the stools. In many cases known to be heavily infected with bilharzia, there are periods in which no ova can be found, and on account of this fact, amply demonstrated in several of our cases of bilharziosis, we conclude that we have passed over many such in our daily clinic."

the present writer feels that the honor of first studying intestinal schistosomiasis in the indigenous population of the Western Hemisphere without question belongs to González Martínez, since his publication antedated that of Letulle<sup>2</sup> (May, 1904), in which a case of rectal schistosomiasis, contracted in Martinique, was reported.

In rapid succession numerous findings of intestinal schistosomiasis, in which lateral-spined eggs were recovered in the feces and in which the urine was negative for eggs, appeared in the literature for the Caribbean area and slightly later for South America. The positive records of McDonell for Culebra and of Holcomb<sup>3</sup> for Vieques, both small islands off the east coast of, and belonging to, Puerto Rico, were reported to the Surgeon-General of the U. S. Navy and appeared in his annual report for 1906 (p. 102). Likewise Gunn<sup>4</sup> had found lateral-spined eggs of schistosomes in the feces of Puertorricans as far away as San Francisco. Thus by 1907 the distribution in the Western Hemisphere of indigenous-intestinal schistosomiasis, uncomplicated with the vesical type, included the following locations: Antigua, Puerto Rico, Martinique, Santo Domingo, Barbadoes, Venezuela and Brazil. Since that date other islands of the Antilles and the Guianas have been involved. The reports for cases from the Canal Zone, Colombia, Jamaica and Costa Rica, all appear to be based, not on indigenous cases, but only on patients who had contracted the disease elsewhere and had been diagnosed there.

In 1907 Sambon<sup>5</sup> named the schistosome, which had lateral-spined eggs that were passed only in the feces, *Schistosoma mansoni*, in honor of Sir Patrick Manson. The morphological studies of Pirajá da Silva<sup>6</sup>, the pathological investigations of Flu<sup>7</sup> and of Risquez<sup>8</sup>, and the life history studies of Leiper<sup>9</sup> provided ample justification for the hypothesis of the pioneers, that the schistosome producing lateral-spined eggs and involving the inferior mesenteric veins as the primary seat of residence, was, indeed, a distinct species.

Following up his earlier work, González Martínez<sup>10</sup> (1916) published an extensive report of his studies on the prevalence and clinical aspects of schistosomiasis mansoni in Puerto Rico. Among other information this contribution indicated that the infestation was prevalent around the entire coast of the Island, wherever cane was cultivated, and in the valleys

crossed by large rivers; that the disease had no predilection for sex, age or race; that the majority of cases acquired the infection through exposure of the skin, but that some cases must have inoculated themselves by way of the alimentary tract; that in certain regions, as in Mayagüez, the incidence was as high as 8.4 per cent, and that in light infections ("benign infections") at least ten years is required after leaving an endemic area before a spontaneous cure can be affected. A few years later this report was followed by a monograph<sup>14</sup> on the same subject, and a masterly and comprehensive contribution on intestinal schistosomiasis<sup>15</sup>.

With the opening of the School of Tropical Medicine at San Juan in 1926 there was renewed interest in schistosomiasis mansoni in the Island. Studies on the epidemiology and distribution of the disease and experimental proof that *Planorbina guadeloupenensis* was the intermediate host of the organism in Puerto Rico were published by Hoffman<sup>10</sup>. Certain other species of cercariae found in *P. guadeloupenensis* were studied by Marín<sup>11</sup>. The pathological anatomy of the disease, as determined by human autopsies and animal infection, was investigated by Lambert<sup>12</sup> and by Lambert and Burke<sup>13</sup>. Also immunological studies were reported by Taliaferro, Hoffman and Cook<sup>14</sup>. Meanwhile González Martínez<sup>15</sup>, in summarizing his contributions to the subject, indicated that he had on circumstantial evidence suspected *Planorbina guadeloupenensis* as the intermediate host of *Schistosoma mansoni* as early as 1921, and advanced evidence in support of the view that the lesions produced by the worm in man have a three-fold origin, namely, "the irritative action of the egg; the sclerosis produced by the chemical action of a phlogogenic toxin secreted by the adult parasite and by its ciliated embryo; and, finally, the endovascular traumatic effects of the adult parasites, producing not a thrombotic but a vegetative endophlebitis, pathognomonic of our schistosomiasis."

The pathological report for the first three years of the School of Tropical Medicine (1926-1929) showed that out of 225 autopsies performed, mostly from the vicinity of San Juan, 30 or 13.3 per cent showed the presence of *Schistosoma mansoni* infection (Burke<sup>15</sup>). Of the 144 autopsies performed during the next fourteen months 10 harbored *S. mansoni*. In commenting on these cases Koppisch<sup>16a</sup> re-

marked: "Of the 10 cases of schistosomiasis, eight presented pseudotubercles in the various organs, mostly liver, in addition to changes in the lower gut. In some they have been found in the pancreas, spleen, suprarenal glands and lungs. Fragments of ova formed the nucleus of the pseudotubercles. One case, that of a young man of sixteen, presented an extensive colloid carcinoma of the cecum with metastases to the peritoneum. Schistosome ova were present in the tumor tissue. In two of the cases the bilharzia ova were found only in the feces."

In 1931 Taliaferro and Taliaferro<sup>17</sup> published information on the skin reactions of *Schistosoma mansoni* cases, using as an antigen extract of dried *Planorbina guadeloupensis* livers infested with *S. mansoni*. In testing 53 persons "with a known present or past infection, 40 were positive, 6 were negative and 7 were eliminated because of positive controls" (i. e. reaction to extract of uninfected snails of the same species); whereas "in tests on 46 uninfected persons, none were positive, 42 were negative and 4 had to be eliminated because of positive controls."

In 1930, Serra<sup>18</sup> reported the results of a survey for helminths based on 2,200 fecal examinations made in the southern and western parts of the Island, particularly from Guayama, Ponce and Mayagüez. Of 2,000 samples from urban centers, 7.35 per cent were positive for *Schistosoma mansoni* (1,000 males, 10.7 per cent, 1,000 females, 4.0 per cent). In the Guayama series (472 males, 444 females) 17.2 per cent of the males and 9.0 per cent of the females were involved. In Ponce only 5 cases out of 679 were found to be positive. In Mayagüez no females and 9.4 per cent of the males were infected. In the heavily infected Guayama area all of the positive females were between 5 and 20 years of age, while 88 per cent of the males were found within this age group.

Aside from the clinical data published by González Martínez this very important aspect of the disease appears to have received little attention in Puerto Rico. In 1922 Carrasquillo<sup>19</sup> reported briefly on the use of tartar emetic in the treatment of schistosomiasis mansoni. In 1928 Pila<sup>20</sup> published a case history of schistosomiasis involving the fallopian tube and the ovary. In 1930 Ramón Suárez<sup>21</sup> presented information on a case with a high eosinophilia (ranging

from 62 to 74 per cent), with ova of *S. mansoni* in the stools and with pulmonary symptoms simulating bronchial asthma, but without appreciable abdominal symptoms. The following year Bonelli<sup>22</sup> wrote of his observations on late schistosomiasis with hepatic cirrhosis, splenomegaly, ascites, hematemesis and leukopenia, and suggested that this phase of the disease was analogous to Banti's syndrome. In commenting on one of these cases which came to autopsy Koppisch<sup>10b</sup> stressed the need for careful differential diagnosis of advanced cases of schistosomiasis mansoni resembling Banti's disease. Gould<sup>23</sup> has reported, in a patient without patent symptoms of schistosomiasis mansoni, suffering from periapendicular abscess, the presence of *Schistosoma mansoni* eggs infiltrated into the involved tissues. Pons and Hoffman<sup>24</sup> have studied the febrile phenomena in a group of carefully selected cases of the disease and have demonstrated the need for differential diagnoses in this type of pyrexia.

#### THE PRESENT INVESTIGATIONS

It has been realized that many of the perplexing problems associated with schistosomiasis mansoni could probably be elucidated by an intensive investigation comprehending all of the various aspects of the disease. The need for such a study and the reasons why Puerto Rico is nearly ideal for the work has recently been presented (Faust<sup>25</sup>). Progress in the investigation has thus far proceeded so satisfactorily as to justify the presentation of an outline of the several phases under consideration. Each chapter will be complete in itself, but will be intimately related to the preceding and succeeding chapters. It is planned to publish the series, of which the foregoing article is the first, in consecutive numbers of this Journal.

#### STUDIES ON SCHISTOSOMIASIS MANSONI IN PUERTO RICO

- I. The history of schistosomiasis mansoni in Puerto Rico.
- II. The epidemiology and geographical distribution of schistosomiasis mansoni in Puerto Rico.
  1. Epidemiology of the infection on the Island.
  2. Parasitological surveys in endemic areas.
- III. Biological studies on schistosomiasis mansoni in Puerto Rico.
  1. The extra-mammalian phase of the life cycle.
  2. The mammalian phase of the life cycle.
- IV. Host tissue reactions in experimental schistosomiasis mansoni.

- V. Clinical studies on schistosomiasis mansoni.  
 1. Clinical field studies.  
 2. The course of the disease.
- VI. The pathology of the disease in man.
- VII. Preventive aspects of schistosomiasis mansoni in Puerto Rico.

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