SCHISTOSOMIASIS IN A CYSTIC TERATOMA OF THE OVARY

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Summary: The authors report the case of an ovarian teratoma infected by the eggs of the trematode *Schistosoma haematobium*. In consideration of the rare observation, bearing a paradigmatic value, they briefly discuss the particular tropism that the parasite demonstrated towards some tumoral structures and they also outline the immunological mechanisms activated by the Schistosoma.

INTRODUCTION

Since the first observation of the ovarian bilharziasis by Symmers [1] in 1906, a lot of reports concerning either localized ovarian infection or generalized involvement of the female genital tract have been made. These cases were mostly due to a *Schistosoma mansoni* (SM), seldom to a *Schistosoma haematobium* (SH) or *Schistosoma japonicum*. The whole subject has been thoroughly revised by Tiboldi [2] who collected the entire literature in an extensive monograph. Nevertheless, even the association between bilharziasis and ovarian tumors has been thoroughly discussed. The first case was reported by Werneck and Junqueira [3] (association between bilharziasis and bilateral ovarian cystoadenoma). Later on, parasitic eggs were observed in arrhenoblastomas [4, 5], cystoma and cystic teratomas [2, 6], granulosa cell tumor and carcinoma [7], dysgerminoma [8], and Brenner's tumor [9]. The adult SM worm was also found within the vessels of an ovarian cystic teratoma [10].

Our case, which is interestingly similar to the one reported by Paradinas [6], could represent a further new contribution to the knowledge of the pathophysiology of Schistosoma infection.

CASE REPORT

A 19-year-old Somalian woman, born in Kisimajo (Low Giuba, Somalia), was admitted to Banadir Hospital, Mogadiscio, complaining of abdominal enlargement. Past history was unremarkable. She referred menarche at age 14; menses followed regularly for rhythm, quantity, and duration. Six months before the admission, she got married. At that time she noted a slow and progressive swelling of the abdomen: as she believed to be pregnant, she didn't worry at first, until she realized the persistence of normal menstrual cycles.

On admission, an abdominal mass was appreciated by physical examination, mostly palpable in the left pelvis. Ultrasound scanning showed that the mass belonged to the left ovary and was partially connected to the right one. She underwent laparotomy according to Pfannenstiel; a resection of the tumor was done, and both the fallopian tubes and the right ovary were preserved. This latter was micropolycystic in appearance.

At gross examination, the tumor was spheroid, whitish with translucent areas, with a clenched-fist-like surface. It largest diameter was 16 cm. Cut surface disclosed several irregular cystic spaces, filled with a clear liquid, mucus, and a chalky-white substance.

Histological examination showed an odd and complex mixture of solid and cystic areas. The former were composed of nerves, ganglia, vascular structures, fibrous and myxoid tissue. The latter displayed a similar heterogeneity and, even more, had a peculiar characteristic: it was composed of numerous eyeball buds, sometimes with a well differentiated retina. There were

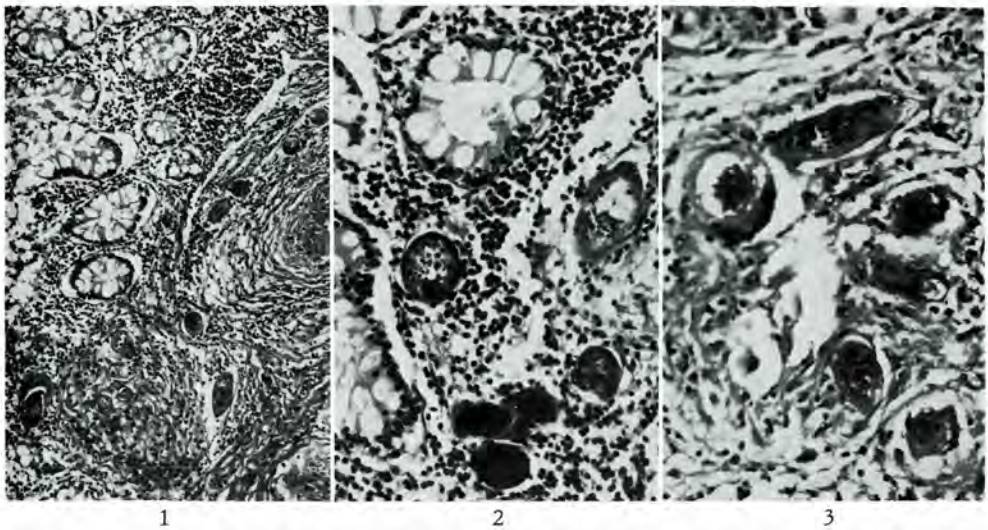

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Fig. 1. — SH eggs in the wall of a cyst lined by an intestinal-type epithelium. Inflammatory reaction is mostly granulomatous, composed of lymphocytes, plasmacells, and foreign-body giant cells (Hematoxylin and eosin, 16 ×). Fig. 2: SH eggs showing incomplete calcification (Hematoxylin and eosin, 40 ×). Fig. 3: SH eggs showing degenerative facts. The typical hook can be recognized (Hematoxylin and eosin, 100 ×).

also other cysts, lined by squamous, ciliate, or intestinal-type epithelium (fig. 1). Within the walls of some cysts lined by intestinal epithelium, several SH eggs could be recognized. Many of them bore the typical hook and were surrounded by a granulomatous reaction, with a few lymphocytes, plasma cells, numerous giant cells, of the foreign-body type (fig. 2 and 3). Phagocytosis of eggs by giant cells was a prominent feature. Some eggs showed regressive factors, like calcification (fig. 2). Adult flukes were not observed in any histological section of the tumor.

After surgery, the patient recovered completely. Further investigations for both SH eggs and hooks in the uterine and stools, performed from a few days after the operation up to four weeks later, gave negative results.

DISCUSSION

Besides the objective rarity of the finding ([2]), the present case report bears some distinctive features which can be summarized as follows:

1) Schistosoma tendency to locate within peculiar structures of the tumor;

2) pattern of inflammatory reaction against the parasite.

As far as the first point is concerned, we have observed SH eggs exclusively within the cystic walls lined by an intestinal-type epithelium, that is, in a structure which recalls the normal mucosa and submucosa of the large bowel. No egg was detected anywhere else, especially in the other numerous squamous and eyeball-bud-like cysts. As a matter of fact, these findings could suggest a selective behaviour of the parasite, rather than the result of a "random" localization. Supporting evidence to such hypothesis has been given by several authors who documented that both the eggs and the adult, worms of S can reach the ovary through port-caval anastomoses ([11, 12, 13]), and that flukes can grow in cystic teratomas ([10]); nevertheless, the ovarian infection by S is still a rare event.

As far the second, namely the host reaction against the parasite, we should re-

member that this subject has still to be explained and is rather a moot point. Berry ([14]) disclosed a mild reaction among ten cases of ovarian bilharziasis due to SH; Paradinas ([6]) also found a mild inflammation in one case of ovarian teratoma. Other authors reported such a marked granulomatous reaction, that they appropriately labelled these cases "granulomatous ovarites" ([15]).

As a matter of fact, our case shares several characteristics with the one reported by Paradinas ([6]). We also agree with Paradinas on the degree of inflammation as being closely related to the duration of the infection, as well as calcific eggs as acting like a mild antigenic stimulus. Last but not least, we ought to remark the bizarre response of the immunitary system against a parasite localized in a tumor; in other words — despite the infection interested an ovarian teratoma, by definition a non-self structure — the immune response acted as if it were protecting a self structure, thus defending the neoplasm.

ACKNOWLEDGEMENTS

The Authors thank prof. Domenico Pecorari for having kindly revised the text.

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