

# Hepatic Schistosomiasis: Report of a Case Accompanying a Metastatic Tumor in the Liver

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Schistosomiasis is secondary to malaria in its frequency among parasitic infections in the third world countries. It is estimated that 200 million people worldwide are infected with schistosomiasis. Five species of *Schistosoma* which are *S. mansoni*, *S. japonicum*, *S. mekongi*, *S. intercalatum* and *S. hematobium* are known to cause infection in man (1). The worldwide distribution of Schistosomiasis is restricted to some tropical and subtropical regions of the world that does not cover Turkey. A few cases which have been reported in our country were found to have visited endemic regions in the past (2). *S. mansoni* is prevalent throughout Middle East, Saudi Arabia, Egypt and Africa as well as South America and Caribbean Islands while *S. japonicum* is mostly found in Far East, Japan and China *S. mekongi* is endemic in South Asia and *S. intercalatum* is found in central Africa. Ten percent of the cases in these endemic regions has hepatosplenic disease (1). Hepatosplenic Schistosomiasis is a manifestation of infections with *S. mansoni*, *S. japonicum* and *S. mekongi* (3). The relation of *Schistosoma* infection with hepatocellular, colonic and bladder carcinomas is still debated (1,4,5). A very rare infection of *Schistosoma* accompanying a metastatic carcinoma in the liver and diagnosed incidentally during the evaluation of liver biopsy performed for the metastatic lesion, is presented in this report.

## Case report

Sixty seven years old male patient living in İzmir presented with a vague right upper quadrant pain. On physical examination, hepatomegaly was detected and abdominal ultrasound was performed, which revealed multiple masses with the largest diameter of 4cm in the liver, raising the possibility of metastatic disease. Laboratory tests revealed elevated ALT, AST, alkaline phosphatase and  $\gamma$ -glutamyl transpeptidase levels. Thoracic X-ray revealed plumpness on left hilar area, while endoscopic examination of the stomach and colonic X-ray showed no mass lesion. Ultrasound guided liver biopsy was performed for histopathologic diagnosis. On light microscopic examination, 3x2x2mm of liver biopsy material was found to be completely composed of tumor tissue. Tumor was composed of solid nests of cells with atypical hyperchromatic nuclei with focal molding effect and scanty cytoplasm consistent with small cell carcinoma (Figure 1). At one edge of the tumor tissue was an oval shaped

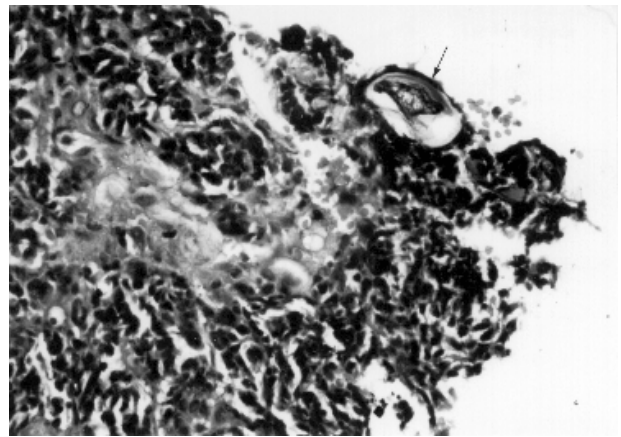


Figure 1. *Schistosoma* egg (arrow) within the tumor tissue.

egg with lateral spine measuring about 150 microns consistent with the definition of egg of *S. mansoni* (Figure 1). There was no recognizable tissue reaction like granulomas around the egg. The stool test, which was performed twice, was negative for ova and parasites. On further examination of the primary site, thorax computerized tomography revealed a mass next to the left main bronchus. Histopathologic evaluation of bronchoscopic aspirate confirmed the diagnosis of small cell carcinoma of the lung. The patient had a family history of lung cancer and suffered malaria when he was young. He had not been to an endemic place of schistosomiasis. He had only been to Korea, which to our knowledge, is not an endemic region for schistosomiasis, during the Korean War. The patient was treated with chemotherapy for his primary tumor. On the course of his therapy, disseminated bone and brain metastases occurred and the patient died about 1 year after the final diagnosis. Autopsy was not performed.

## Discussion

Among the human diseases that are found endemically in different parts of the world, schistosomiasis is one of the most extensively studied infection, not only because of the morbidity and the mortality it produces but also because of the large number of infested people worldwide (6).

Neither Turkey nor Korea in which the patient lived during his life time is included in the list of the countries which are endemic for schistosomiasis. This case may imply that research about epidemiology of schistosomiasis should be widened. In endemic areas, 10% of the

patients with schistosomiasis develop hepatosplenic involvement. Hepatosplenic schistosomiasis results from infection with mansoni which has a similar life cycle like any other schistosoma species.

*S. mansoni* eggs which then free a larva called miracidium are found in fresh water. Miracidium enters a snail and develops into cercaria, which is then released into the water again to penetrate the skin. Adult forms of schistosomes are found in mesenteric vascular bed where they lay eggs. Some eggs are trapped in the wall of the viscus and others are taken with the flow of the portal vein to the liver. Eggs which are trapped in the intestinal wall migrate to the lumen and are eliminated by stool. On the other hand, eggs which are trapped in the portal venules and reach the portal areas of liver, measure 116 to 180 microns and have a large distinctive lateral spine that protrudes from the side of the egg near one end (7). The initial reaction is an immune response to egg antigens which consequently results in histopathologic findings (7,8). On microscopic examination of liver biopsy infected with schistosomes, ova are seen in the portal and periportal regions. Portal tracts show varying amounts of fibrosis and generally contain an inflammatory infiltrate which often includes eosinophils. Schistosomal pigment may be present resembling malarial haemozoin. Lobular architecture of the liver is essentially maintained. In schistosomiasis, although serious morbidity and even death result from tissue fibrosis following granulomatous inflammation; the mechanism involved in the pathogenesis of fibrosis remains uncertain (1,3). In a study of Prakash et al, it was concluded that T cells influence the production of fibrogenic factors produced by granulomas (9). The liver is usually found to be enlarged although it may be normal or reduced in size.

In our case, secondary changes due to the egg could not be seen as the whole tissue was composed of tumor. The typical histological appearance of the egg has led us to the diagnosis of schistosomiasis. Besides, no clinical signs of portal hypertension could be seen. Although our patient had some vague gastrointestinal complaints before, he had no stigmata of chronic liver disease.

Diagnosis of schistosomal infection is definite only when eggs of the parasite are demonstrated in stool samples or tissue biopsies like rectal mucosa (1). Liver biopsies can also demonstrate eggs as in our case, but often can miss the lesion. Watt et al have derived a reasonable algorithm for the physicians considering the diagnosis of hepatosplenic schistosomiasis recommending collection of 5 stool samples and serum for serologic tests. If these tests are negative, rectal biopsy should be obtained (1). In our patient, schistosomiasis had not been suspected

before the diagnosis with liver biopsy, therefore no serological tests were done. After the diagnosis, stool examination was performed twice but no parasites could be demonstrated.

There have been numerous attempts to establish an etiologic relationship between hepatic schistosomiasis and liver cancer. Although, in some studies, the incidence of hepatocellular carcinoma in patients with chronic hepatic schistosomiasis has been found to be higher compared to those without parasitosis, serologic evidence of HBV infection was present in most of the cases (1). These data might suggest that schistosomiasis and Hepatitis B infection may be synergistic, therefore direct relationship between parasitic liver disease and hepatocellular carcinoma remains to be proven.

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