

# Splenosis in patient with *Schistosoma Mansoni* infection who underwent splenectomy and auto implantation of spleen morsels in the major omentum 37 years ago: Case report

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## Abstract

**Introduction:** Splenosis has been widely reported around the world with an incidence of 16-67% after traumatic splenic rupture or splenectomy, however, long term follow-up of intentional splenosis after splenectomy, ligature of the left gastric vein and auto implantation of spleen morsel in the major omentum for portal hypertension in children due to mansonic schistosomiasis (MS) is rare or not yet published. The purpose of this report is the presentation of a patient with patent splenosis in abdominal surgery for pain with the diagnosis of tail pancreas tumor. This is the first report of long-term follow-up (more than 37 years) from a child who underwent splenectomy and auto implantation off spleen morsels in the major omentum.

**Case report:** VL, 39-year-old, born in the north eastern region of Brazil (hyperendemic area for schistosomiasis mansoni), underwent laparotomy for abdominal pain with the diagnosis of a pancreatic mass in its tail. The preoperative image investigation (computed tomography) showed several nodules in the upper portion of abdomen and a mass in the tail of the pancreas with close relationship with the stomach. Presumptive diagnosis was pancreatic tumor (Frantz's tumor). She underwent laparotomy partial resection of the pancreas with removal of the mass, which was sent for histology. It was observed the intentional splenosis, which was anatomically very good (resembling an accessory spleen), which was not involved in the pancreas mass. Her post-operative was with no complication. In the long past (childhood) she had had splenectomy, ligature of the left gastric vein and auto implantation of spleen morsels in the major omentum when she was 13-year-old, due to esophageal hemorrhage caused by portal hypertension due to hepatosplenic Schistosomiasis mansoni. At that time, she also had hypersplenism and low body development. After surgery, she grew up as normal adolescent with no esophageal bleeding and no repeated infection nor overwhelming post splenectomy infection (OPSI). She married and gave birth of a normal child who is now 14-year-old.

**Comments:** This is a very successful case of auto implantation of spleen morsels in the major omentum, after splenectomy and ligature of the left gastric vein, in a patient with, at childhood, severe esophageal bleeding due to portal hypertension caused by hepatosplenic schistosomiasis mansoni. The reported case highlights the importance of maintaining some spleen tissue after splenectomy, in children, after spleen removal either due to trauma or benign disease, as it was the present report. The relationship between hepatosplenic schistosomiasis mansoni and cancer has been reported and it is under investigation.

**Abbreviations:** OPSI: Overwhelming Post Splenectomy Infection; MS: Mansonic Schistosomiasis; SM: Schistosomiasis Mansoni.

## Introduction

Schistosomiasis mansoni is a chronic liver disease, in which some patients progress to the most severe form (5% to 7%), hepatosplenic schistosomiasis. This form is associated with portal hypertension, splenomegaly, and often episodes of upper digestive tract bleeding, even with liver function preserved [1-4].

Splenectomy and ligature of the left gastric vein, gastric devascularization or distal splenorenal shunt are validated procedures to reduce portal hypertension following digestive bleeding in these patients [5-8]. In children, autoimplantation of spleen morsels into the major omentum is mandatory [9].

There are few reports on the fate of spleen morsels auto implantation after splenectomy in patients with portal hypertension due to schistosomiasis mansoni. Furthermore, this mean late follow-up is  $12.1 \pm 5.6$  years. To the best of our knowledge this is the first report of a patient with esophageal bleeding due to schistosomiasis mansoni who underwent splenectomy and spleen morsels auto implantation after 37 postoperative years of the original surgical procedure. Thus, being the purpose of the scientific communication.

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## Case report

VL, 39-year-old, born in the north eastern region of Brazil (hyperendemic area for schistosomiasis mansoni), underwent laparotomy for abdominal pain with the diagnosis of a pancreatic mass in its tail. The preoperative image investigation (computed tomography) showed several nodules in the upper portion of abdomen (Figure 1) and a mass in the tail of the pancreas with close relationship with the stomach.

Presumptive diagnosis was pancreatic tumor (Frantz's tumor). She underwent laparotomy partial resection of the pancreas with removal of the mass, (Figure 2) which was sent for histology.

It was observed the intentional splenosis, which was anatomically very good (resembling an accessory spleen), which was not involved in the pancreas mass (Figure 3).

Apart from surgical wound infection, her post-operative was with no complication. In the long past (childhood) she had had splenectomy, ligation of the left gastric vein and auto implantation of spleen morsels in the major omentum when she was 13-year-old, due to esophageal hemorrhage caused by portal hypertension due to hepatosplenic *Schistosomiasis mansoni*. At that time, she also had hypersplenism and low body development. After surgery, she grew up as normal adolescent with no esophageal bleeding and no repeated infection nor overwhelming post splenectomy infection (OPSI). She married and gave birth of a normal child who is now 14-year-old. In the pre-operative period she was assessed for blood cell count, especially for lymphocyte blood and immunoglobulin levels, which revealed: absolute lymphocyte count  $1,484/\text{mm}^3$ ; total T absolute lymphocyte  $\text{CD3}^+ 831/\text{mm}^3$  - 56%; T auxiliary lymphocyte  $\text{CD4} (\text{CD3}+\text{CD4}^+) 490/\text{mm}^3$  - 33%; T suppressor lymphocyte  $\text{CD8} (\text{CD3}+\text{CD8}^+) 282/\text{mm}^3$  - 19%;  $\text{CD4}/\text{CD8}$  ratio 1.74; lymphocyte B  $\text{CD19}^+ 178/\text{mm}^3$  - 12%; sub population  $\text{CD3}^-/\text{CD16}^+/\text{CD56}^+ 312/\text{mm}^3$ . The immunoglobulin assessment showed: rubella IgG antibody - 113.0 UI/ml, rubella IgM antibody - 127 mg/100ml; total IgE 66IU/ml; IgG -1,471 mg/100ml; IGA - 302 mg/100ml. Although  $\text{CD4} (\text{CD3}+\text{CD4}^+) 490/\text{mm}^3$  was at the level of immunosuppression, all other lymphocyte counts, and immunoglobulin levels were within the reference confident interval for her age. She presented with no signs of hypersplenism.

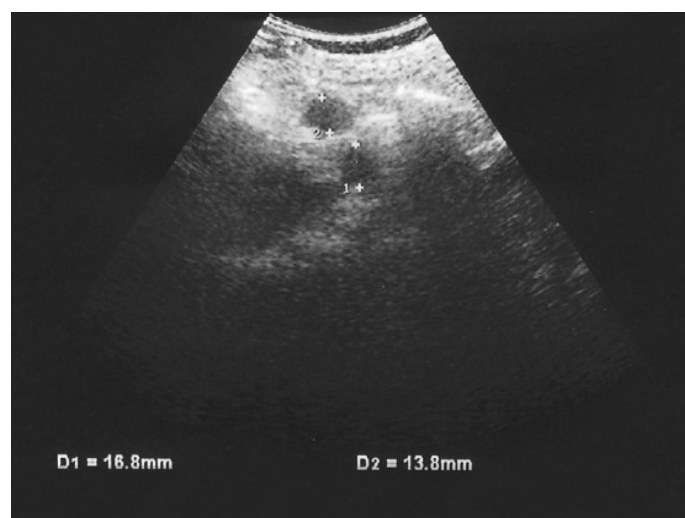


Figure 1. Spleen nodules seen on ultrasound

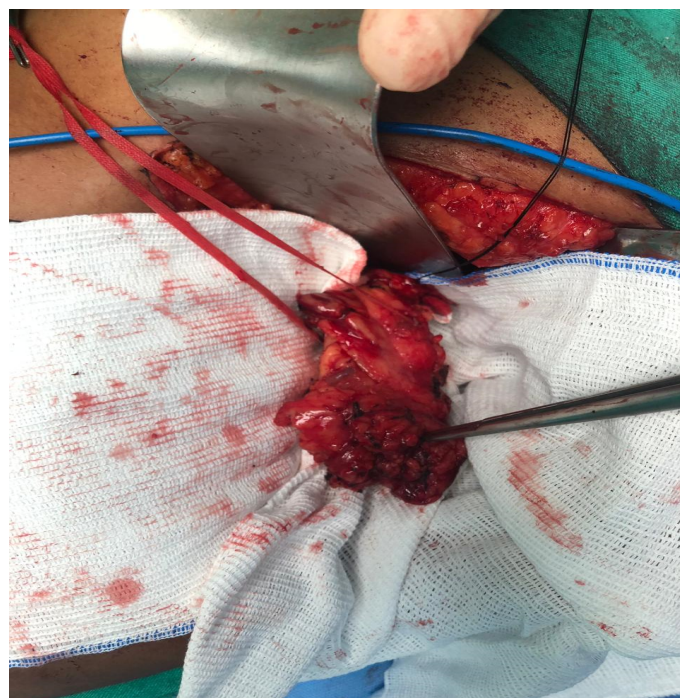


Figure 2. Surgical specimen of the pancreas tail and the tumor



Figure 3. Spleen nodule, resembling an accessory spleen, just below the greater gastric curvature. A site where a spleen morsel was implanted 37 years ago

## Comments

The schistosomiasis mansoni (SM) is endemic in the northeastern region of Brazil, and its main factor of morbidity is the portal hypertension, which occurs in about 5% of the patients. In the severe cases of bleeding from esophageal varicose veins, surgery is the treatment for portal decompression, generally including splenectomy, which reduces portal hypertension decreasing the risk of posterior bleeding [1,5-10]. Auto implantation of spleen morsels in the major omentum has the potential to reduce the risk of overwhelming postsplenectomy infection (OPSI) [9,10].

The proved splenosis, in this patient, was surrounded by splenic capsule (accessory spleen). This is a very interesting finding because the spleen morsels auto implanted thirty-seven years ago had no capsule, which raises the question of splenic regeneration. Until now, spleen functionality, in splenosis, is assessed by splenic regeneration. However, the issue still remains an interesting pathological aside whose benefit to patients remains an enigma and societal value unexploited [11,12].

The peripheral blood cell count of this patient was within the normality, which was interpreted as she cured of her hypersplenism that existed previous to original surgery and she was in hematologic good condition for undergoing major surgery.

As regard to low CD4 lymphocyte in this patient, it should not be taken as exclusion criteria for surgical intervention as it not happens in cancer and HIV/AIDS patients [13,14]. The management of immunodeficiency in surgical patients and the use of anesthetic substances need further study [13]. Furthermore, surgical procedures in HIV/AIDS patients require good expertise both in surgery and in infectology [13,14].

The relationship between hepatosplenic schistosomiasis and cancer has been investigate and, equally, requires further studies [15,16]. The present report should be taken into consideration for future epidemiological investigations on this subject especially regarded to immunology.

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