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Case Report

Extrahepatic portal hypertension due to chronic portal vein thrombosis with cavernous transformation treated with distal splenorenal shunt and postoperative massive lymphocele: a case report

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ABSTRACT

Extrahepatic portal hypertension in childhood is an important condition, frequently caused by chronic portal vein thrombosis with cavernous transformation. When physiological reconstruction is not viable, the selective portosystemic shunts remain as a useful option to preserve liver function. It is presented the case of a 13-year-old male adolescent with extrahepatic portal hypertension secondary to portal cavernoma. He had antecedents of upper gastrointestinal bleeding and severe hypersplenism. Because the portal anatomy was not suitable for a meso-Rex bypass, it was decided to perform a distal splenorenal shunt. In the postoperative period, the patient presented a massive abdominal lymphocele of high volume. The management was conservative, placing a closed Blake drainage and initiating total parenteral nutrition with fasting. After these measures, the lymphatic output had a progressive reduction until its total resolution, without needing more invasive surgeries. The distal splenorenal shunt is a valid alternative in patients with preserved liver function that cannot receive a meso-Rex bypass. The postoperative lymphocele is a rare complication in this surgery, but its conservative management with close observation can be sufficient and successful.

Keywords: Extrahepatic portal hypertension, Portal vein thrombosis, Portal cavernoma, Distal splenorenal shunt, Lymphocele

INTRODUCTION

Portal hypertension in childhood is less frequent than in adults, but its impact is considerable due to the risk of upper gastrointestinal bleeding, hypersplenism and possible repercussions on growth and quality of life. According to Grama et al, chronic portal vein thrombosis with cavernous transformation is one of the main causes of extrahepatic portal hypertension in children and it represents around 30–40% of cases.

In many of these patients the hepatic parenchyma remains structurally preserved, which allows considering surgical strategies aimed at decompressing the portal system while attempting to preserve liver function.[1]

The factors associated with portal thrombosis in pediatric age are; history of umbilical catheter, neonatal sepsis, hereditary or acquired prothrombotic states and abdominal inflammatory processes. However, in a proportion of children a specific cause is not identified, as reported by Shneider et al and the guidelines proposed by Flores-Calderón et al for extrahepatic portal obstruction, this variability in etiology requires extensive clinical and imaging evaluation. In practice, the most common presentation remains the combination of splenomegaly, thrombocytopenia, and esophageal varices in a patient without a history of chronic liver disease.[1,2,10,11]

The initial management of extrahepatic portal hypertension includes pharmacological prophylaxis with non-selective beta-blockers and endoscopic therapy for

varices.[2,7,10] When despite these measures there is a persistent high risk of bleeding, significant hypersplenism remains or quality of life is compromised, surgery is considered. The ideal alternative is physiological reconstruction of portal flow, such as the meso-Rex bypass, as long as the intrahepatic and mesenteric portal anatomy allows it.[6,12] However, in patients with extensive thrombosis of the main portal vein with cavernous transformation physiological reconstruction may not be viable. In that context, selective portosystemic shunts, particularly the distal splenorenal shunt (Warren surgery), remain useful in decompressing the splenoportal territory and preserving part of the hepatic portal flow, with good results in controlling bleeding in selected series of pediatric populations.[8,9,12,15]

Complications of this type of shunt include shunt thrombosis, infections, chylous ascites, encephalopathy (less frequent with a healthy liver), and lymphatic collections.[3,8] Postoperative abdominal lymphocele is a rare complication in pediatric surgery; it has been reported more frequently after retroperitoneal lymphadenectomies, vascular procedures, and transplants, and its management is usually stepwise: drainage, modified diet, minimally invasive approaches, and even surgery in certain cases.[3,13,14] Due to the scarcity of reports in children undergoing distal splenorenal shunt, detailed documentation of these cases provides information to recognize these complications earlier and adjust therapeutics.

CASE REPORT

A 13-year-old male adolescent with a diagnosis since childhood of extrahepatic portal hypertension secondary to chronic portal vein thrombosis with cavernomatous transformation, history of several episodes of upper gastrointestinal bleeding due to esophageal varices treated with endoscopic ligation and management with propranolol. Neonatal history of perinatal asphyxia, meconium aspiration syndrome and umbilical catheter use. No previous abdominal surgeries, chronic liver disease or coagulation disorders were documented. He attends the referral hospital for scheduled evaluation due to persistence of marked splenomegaly, severe thrombocytopenia, and high risk of variceal re-bleeding despite medical and endoscopic management. On physical examination upon admission, he is hemodynamically stable with discrete pallor, soft and non-painful abdomen, with evident splenomegaly, without signs of ascites and no frank hepatomegaly.

Preoperative laboratories report: hemoglobin 15 g/dl, hematocrit 44.3%, leukocytes 2.46 K/µl, platelets 31,000/µl, prothrombin time 13.3 s, INR 1.203, aPTT 27.1 s and serum creatinine 0.50 mg/dl. Liver function tests: total proteins 7.9 g/dl, albumin 4.6 g/dl, A/G ratio 1.4, AST 62 U/l, ALT 46 U/l, alkaline phosphatase 326 U/l, total bilirubin 1.97 mg/dl (direct 0.47 mg/dl and indirect 1.50 mg/dl). These findings suggest preserved liver and renal function, with cytopenias mainly attributable to hypersplenism, in accordance to what is described in series of pediatric portal hypertension.[1,2,11,12]

Hepatic Doppler ultrasound shows a portal vein with an unusual form, multiple periportal collaterals compatible with cavernous transformation, preserved hepatic flow in intrahepatic branches, patent splenic vein and splenomegaly. Contrast-enhanced computed tomography confirms chronic portal vein thrombosis with cavernomatous transformation, splenomegaly occupying more than half of the abdomen, portosystemic collateral circulation and absence of signs of cirrhosis or liver disease. These findings, together with the complex portal anatomy, ruled out the option of a meso-Rex physiological reconstruction and guided us to surgical management through selective portosystemic shunt, in accordance with recommendations in patients for whom direct reconstruction is not feasible (Figure 1).[6,8-10,15]

The portal trunk is replaced by a portal cavernoma, with absence of a suitable intrahepatic portal vein, precluding physiological portal reconstruction by meso-Rex bypass despite a patent superior mesenteric vein. Hemodynamic consequences are dominated by selective splenic venous hypertension, resulting in splenomegaly. Preserved hepatic parenchyma is associated with a low risk of hepatic encephalopathy, supporting a selective portosystemic decompression strategy (Figure 1).

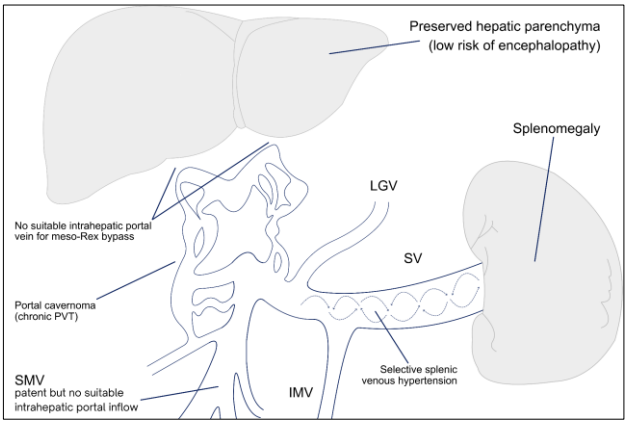

Figure 1: Schematic representation of extrahepatic portal hypertension secondary to chronic portal vein thrombosis with cavernous transformation in the setting of preserved hepatic parenchyma.

Surgical intervention

It was decided to perform a distal splenorenal shunt with the objective of decompressing the splenoportal territory, reducing the risk of variceal hemorrhage, improving hypersplenism and preserving a functional liver.[8,9,12,15]

Under general anesthesia a midline laparotomy was performed. Significant splenomegaly, multiple perigastric and splenic collaterals, and a normal liver in size and

appearance were identified. The splenic vein and the left renal vein were dissected after careful ligation of collaterals and verification of adequate length and caliber. Anastomosis of the splenic vein with the left renal vein was performed with continuous suture while preserving the rest of the available portal flow. No intraoperative complications occurred and the initial shunt patency was considered adequate based on inspection.

Postoperative course and complications

In the immediate postoperative period, the patient was admitted to the pediatric intensive care unit for monitoring. He remained hemodynamically stable without signs of bleeding and with adequate pain control. In the first days a trend toward gradual improvement in thrombocytopenia was observed. However, during the first postoperative week, an increase in abdominal perimeter and drainage output of serous-yellowish fluid in increasing volume was documented. Control abdominal tomography showed a large-volume homogeneous collection adjacent to the shunt path and the splenic hilum without clear communication with the digestive tract, biliary tract or urinary tract. It was compatible with postoperative lymphocele. Liver function tests in this period showed a transient cholestatic pattern with hyperbilirubinemia and moderate liver enzymes elevation, interpreted as post-shunt inflammatory response without signs of frank liver failure.

Given that the patient remained afebrile with stable vital signs and without intense pain, conservative management was decided. A closed Blake drainage was placed and maintained with close monitoring of the volume and characteristics of the fluid. Under this strategy, a gradual decrease in drainage output, improvement in abdominal discomfort and progressive normalization of hepatic parameters in subsequent controls were observed. No sclerosis, surgical marsupialization or additional percutaneous approaches were required. This management is documented in stepwise management schemes proposed for postoperative lymphocele (Figure 2).[3,13,14]

The splenic vein is anastomosed end-to-side to the left renal vein, providing selective decompression of the splenic venous system while preserving hepatopetal mesenteric venous flow. The superior mesenteric vein remains patent and drains toward the portal venous system despite the presence of portal cavernoma. The anatomical relationship between the splenic vein, left renal vein, inferior vena cava, and adjacent retroperitoneal structures is illustrated, highlighting the physiological basis of selective portosystemic shunting in non-cirrhotic portal hypertension. The proximity of the shunt to the left kidney explains the development of a postoperative retroperitoneal lymphocele observed during follow-up (Figure 2).

On the sixth postoperative day, after the start of oral intake, the patient presented a marked increase in Blake drainage output with an approximate volume of 1500 ml/day. Conservative management with octreotide, diuretic, and astringent diet was instituted; however, no improvement was observed with an increase in output up to 2600 ml/24 hours. Given the persistence of high drainage volumes, fasting and initiation of total parenteral nutrition (TPN) were indicated on the tenth postoperative day. After these measures, the output decreased significantly to 200–600 ml/24 hours with progressive reduction until becoming almost null over approximately two weeks, keeping the patient on fasting and with TPN. Once the absence of drainage output was documented, oral feeding was restarted without recurrence of drainage. The diet was scaled progressively and TPN was discontinued on postoperative day 28. No other complications occurred during the evolution.

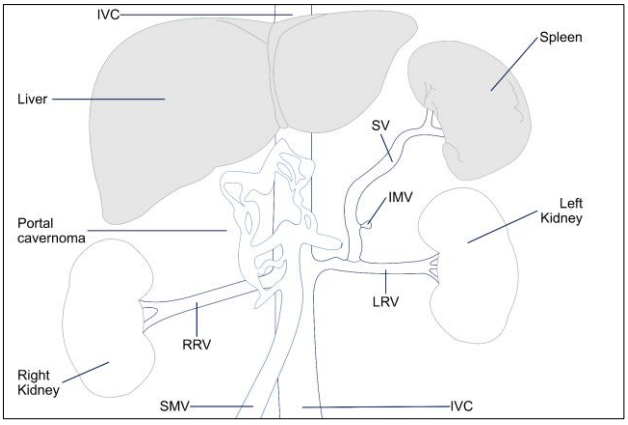

Figure 2: Schematic representation of distal splenorenal (Warren) shunt anatomy following surgical intervention.

Throughout the entire postoperative course no new episodes of gastrointestinal bleeding were recorded, there was no clinical evidence of encephalopathy and no deterioration of renal function. After completing the inpatient monitoring period and documenting clinical stability, adequate oral intake and significant reduction in the volume of the drained lymphocele, the patient was discharged with scheduled follow-up in the outpatient pediatric surgery and gastroenterology clinic for shunt control and definitive resolution of the collection.

DISCUSSION

Chronic portal vein thrombosis with cavernomatous transformation is one of the main causes of extrahepatic portal hypertension in pediatrics and can explain around one-third of cases.[1,10,11] Unlike cirrhosis, in most of these children the hepatic parenchyma is unaltered, which allows considering surgical strategies aimed at decompressing the portal system while maintaining the liver with its function intact.[1,2,7,12] This approach is relevant because it improves the impact on growth and quality of life is important. The ideal management when anatomy allows is physiological reconstruction of portal

flow, with the meso-Rex bypass being the recommended procedure of choice.[2,10,12,15] However, a portion of patients present extensive portal vein thrombosis or complex collaterals, making this procedure unviable. In this context selective portosystemic shunts continue to have a place in controlling variceal hemorrhage and reducing hypersplenism.[8,9] The distal splenorenal shunt described by Warren maintains splenic drainage to the left renal vein and, at least partially, preserves hepatic portal flow through other collaterals with acceptable rates of bleeding control.[8,9,12] Our patient fit precisely in this scenario: anatomy not suitable for meso-Rex, preserved liver function and history of significant bleeding. The development of a postoperative abdominal lymphocele in this case introduces a point of discussion little described in this type of surgery. Lymphatic leak and lymphocele are better recognized in oncological surgeries and extensive lymphadenectomies, with variable incidences but higher than those observed in portal surgery.[3,13] Recent reviews indicate that management should be stepwise: observation, percutaneous or closed drainage, dietary measures, image-guided interventionism and in selected cases surgery.[3,13,14] In our patient, the presence of a large volume collection but good clinical condition allowed opting for a conservative approach with Blake drainage and monitoring, with favorable evolution without the need for additional invasive procedures.

A conservative approach was chosen initially due to suspicion of postoperative lymphatic leak, most postoperative lymphatic collections do not require intervention and tend to resolve spontaneously; so the interventions are reserved for persistent or symptomatic cases. Likewise, simple aspiration was not considered as definitive treatment due to its high reported recurrence and infection and prolonged drainage may be associated with relevant recurrences and extended periods of lymphatic output. In case of persistence of high output, escalation to sclerotherapy through the catheter and/or lymphangiography with Lipiodol with or without lymphatic embolization would have been considered; finally, laparoscopic/open marsupialization would remain as rescue therapy. In our patient, the implementation of fasting and total parenteral nutrition, along with adjuvant therapy, was associated with a sustained decrease in output until its resolution, so escalation to additional percutaneous or surgical interventions was not necessary. This coincides with the recommendation to reserve more aggressive interventions for symptomatic, persistent or complicated lymphocele.[3,13,14]

The relevance of the case lies in several practical aspects. First, it shows the process of selecting surgical technique in extrahepatic portal hypertension when physiological reconstruction is not feasible, supported by imaging and integration of evidence.[6,8-10,15] Second, it demonstrates the utility of monitoring lymphatic complications, which can be confused with biliary or infectious collections and modify the evolution if not identified. Third, it provides experience on the successful conservative management of a lymphocele associated with distal splenorenal shunt in an adolescent, a scenario little reported until now.

Among the limitations of this report are the limited follow-up time to evaluate long-term shunt patency and complete resolution of the lymphocele, which does not allow generalizing results. Nevertheless, by presenting the clinical, surgical, and radiological evolution, we seek to contribute useful information for those facing similar situations.

CONCLUSION

Extrahepatic portal hypertension due to chronic portal vein thrombosis with cavernous transformation continues to be a diagnostic and therapeutic challenge. In patients with preserved hepatic parenchyma, significant variceal bleeding and anatomy not suitable for meso-Rex, distal splenorenal shunt remains a valid alternative.

Postoperative abdominal lymphocele is a rare complication in this context, but possible, and its early recognition allows establishing a proper management. In the presented case, closed drainage and close observation were sufficient, avoiding more invasive procedures that can compromise the shunt objectives.

This case reinforces the need to individualize the surgical strategy in portal hypertension, rely on detailed imaging, and maintain vigilance for uncommon complications.

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