Treatment of Hypersplenism by Partial Splenic Embolization Through Gastric Collaterals

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ABSTRACT

We report a case of Chronic lymphocytic leukemia (CLL) with associated hypersplenism, that was referred to us for partial splenic embolization (PSE) as the patient was not a surgical candidate for splenectomy. Initially, we were not successful in catheterizing the splenic artery from the celiac trunk due to significant atherosclerotic disease. Therefore, we successfully managed to access the distal splenic artery through patent gastro-epiploic collateral circulation along the greater curvature of the stomach. Partial splenic embolization was successfully performed and resulted in improvement of the patient’s peripheral blood cell count as well as 60-70% reduction in the size of the spleen on follow up. Our case highlights an alternative pathway for splenic artery embolization when catheterization of the splenic artery is not feasible. To our knowledge, the use of gastro-epiploic collaterals to embolize the spleen has not been previously reported in literature.

CASE REPORT

A 77 years-old male patient was referred to our service for PSE. The patient was previously diagnosed with CLL 10 years ago. A marked pancytopenia was observed 3 years later. Bone marrow biopsy was obtained, revealing hyperplastic bone marrow which is consistent with the diagnosis of hypersplenism. Hypersplenism is defined as a functional over-activity of the spleen, resulted in peripheral blood cytopenia in the presence of normal or hyperplastic bone marrow cellularity [1]. The patient had a significant past medical history for coronary artery disease and acute myocardial infarction that was treated with coronary stent placement in the left anterior descending artery. He also had atrial fibrillation and underwent cardioversion 5 years ago. The patient had ischemic cardiomyopathy and recurrent left pleural effusion. Given all his co-morbidities, the clinical and anesthesia team decided that the patient is not a candidate for surgical splenectomy, and a decision was made to perform PSE, as a minimally invasive procedure, trying to restore his peripheral blood cell count back to normal.

Computed tomography (CT) scan of the abdomen showed marked splenomegaly with a splenic span of approximately 21 cm, consistent with the patient’s physical examination. The splenic artery appeared severely calcified due to atherosclerotic disease (Fig. 1a). There were multiple hyperdense scattered lesions in the spleen (Fig. 1b) with abdominal and pelvic lymphadenopathy that is related to the patient’s CLL.
The patient gave consent to perform the procedure after being informed of its risks and potential benefits. The right common femoral artery was accessed and initial aortogram showed dilated calcified highly tortuous splenic artery with massive splenomegaly. The celiac artery was then catheterized and celiac angiogram showed moderately dilated gastroduodenal and right gastroepiploic arteries (Fig. 2a). On the delayed images, the right gastroepiploic artery was seen to communicate with the distal splenic artery through its anastomosis with the left gastroepiploic artery at the greater curvature of the stomach (Fig. 2b).

Multiple attempts to catheterize the splenic artery through the celiac artery were unsuccessful given the significant atherosclerotic disease and calcification as well as the severe tortuosity of the splenic artery. Further review of the 3-dimensional volume rendered images of the CT scan, that were performed prior to the procedure, revealed that the anastomosis between the right and left gastroepiploic arteries was patent, and that catheterization of the distal splenic artery from the gastroduodenal artery through the gastroepiploic arteries was feasible (Fig. 3).

The gastroduodenal artery was then catheterized using 2.4 French microcatheter (Progreat; Terumo, Tokyo, Japan). The microcatheter was then advanced through the right gastroepiploic artery and into the left gastroepiploic artery and eventually into the distal splenic artery. Angiogram through the microcatheter confirmed location in the distal splenic artery (Fig. 4). Partial embolization of the lower two-third of the spleen was achieved using 500 um polyvinyl alcohol particles (Embozene; Celonova, San Antonio, Texas). Follow up angiogram through the microcatheter showed complete stasis in the embolized segments of the spleen (Fig. 5).

A broad-spectrum, intravenous antibiotic was prescribed before and after the procedure for a 5-day course. Following the procedure, the patient had left upper quadrant abdominal pain that was scored as 8 on visual analogue scale were 0 represent no pain and 10 represent the most imaginable severe pain. The patient was placed on morphine patient-controlled analgesia pump and his pain gradually improved. The patient was discharged 4 days after the procedure in stable condition.

Continued follow up of the patient over a period of 2 years showed gradual improvement of peripheral blood cell count which returned back to normal levels (red blood cell count went from 1.9 to 4.8 cell/mcl, platelet count went from 67 to 333 cell/mcl). Follow up CT scan done 2 years after the procedure revealed marked decrease (53%) in the splenic volume (2694 cm³ before PVE and 1268 cm³ after PVE) with multiple hypodense areas in its lower two-third which were consistent with multiple splenic infarcts as a result of the prior embolization. The cranio-caudal length of the spleen went down from 22.9 cm to 14.6 cm (Fig. 6).

**DISCUSSION**

**Etiology & Demographics:**
There are several potential causes of splenomegaly and hypersplenism. Nearly half of the CLL patients develop splenomegaly. Occasionally, splenomegaly may present with symptoms of early satiety and/or abdominal fullness. Sometimes, splenic enlargement may result in hypersplenism, contributing to anemia and thrombocytopenia, similar to what was observed in our patient. However, cytopenia may also be due to extensive marrow involvement with CLL and/or intermittent expression of auto-antibodies [1,6-8].

**Clinical findings:**
Some patients may have only mild symptoms of reduced exercise tolerance, fatigue, or malaise. Patients may experience such symptoms even when they apparently lack major organ involvement or anemia. Other patients may present with more advanced disease such as weight loss, recurrent infections, leukemic pleural effusion, bleeding secondary to thrombocytopenia, and symptomatic anemia [2,3]. Because of the advanced age of the affected population, patients sometimes present with an exacerbation of another underlying medical condition, such as pulmonary, cerebrovascular, or coronary artery disease [4,5].

Surgical splenectomy is the definitive treatment for hypersplenism and can eliminate hypersplenism-induced blood cell destruction, but the morbidity and severe complications from surgical splenectomy, including laparoscopic and open splenectomy, still ranges from 9.6% to 26.6% [2,3]. In addition, splenectomy is often associated with an increased long-term risk of septic events [20, 21]. Ligation and banding of the splenic artery are also used to treat hypersplenism, but complications such as portal vein thrombosis, sepsis and multi-organ failure may occur with these methods [13]. Although percutaneous placement of a narrowed stent into the splenic artery is a promising technique for treating hypersplenism [19], it is difficult to ensure a sustained and long-term increase in peripheral blood counts as the splenic artery is not completely occluded, which cannot provide a sustained splenic infarction rate of more than > 50% of the splenic size [22].
Differential Diagnoses:

Causes of splenomegaly and hypersplenism are usually overlapped but attention should be taken as both are different disease entities that might be co-exist in same patient. Splenomegaly is considered when the cranio-caudal length of the spleen exceeds 11 cm. Unlike cross-sectional imaging, ultrasonographic measurement of the cranio-caudal length of the spleen is considered challenging, giving the shape and orientation of the spleen [29,30].

On ultrasonography, the spleen appears as a homogeneous echogenic organ. Normal spleen is slightly more echogenic than healthy liver and markedly hypeerechoic compared to kidneys. The spleen has a homogeneous density in non-enhanced CT scans and usually measures around 45 Hounsfield units [31]. On T1-weighted magnetic resonance images, the spleen shows lower signal intensity and higher signal intensity on T2- weighted images compared to normal liver [32].

Causes of splenomegaly and hypersplenism can be classified into 5 groups:

1. Infectious causes: viral infection like infectious mononucleosis and HIV, bacterial infection as seen in brucellosis and tuberculosis, parasitic infections mainly noted with malaria and visceral leishmaniasis and histoplasmosis as fungal infection.
2. Hyperplastic splenomegaly: which is commonly seen with hereditary spherocytosis, thalassaemia, polycythaemia rubra vera, myelofibrosis, and chronic myeloid leukaemia, chronic lymphocytic leukaemia and lymphoma.
3. Congestive splenomegaly: in cases with liver cirrhosis, hepatic vein obstruction, portal vein obstruction, splenic vein obstruction, congestive heart failure with increased venous pressure, and splenic artery aneurysm.
4. Metabolic and infiltrative splenomegaly: as in cases with Gaucher's disease, amyloidosis, Niemann-pick disease, histiocytoisis, splenic tumors, metastatic malignancy, Marble bone disease and Waldenstrom macroglobulinemia.
5. Miscellaneous causes: Idiopathic non tropical splenomegaly, iron deficiency anemia and B12 deficiency [29] (Table 2 & Table 3).

TEACHING POINT

Marked tortuosity of the splenic artery is a common challenge faced during splenic artery embolization and may result in technical failure of the procedure. We report an alternative novel approach that involves catheterization of the distal splenic artery through gastric collaterals. The technique is an alternative when the standard approach is not feasible.

REFERENCES


**FIGURES**

**Figure 1:** A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: (a) Axial CT image showing significantly enlarged spleen (large arrow) with severe atherosclerotic calcification of the splenic artery (small arrows). (b) Coronal reformatted CT image showing significantly enlarged spleen extending into the pelvis (large arrow) with a hyperdense lesion in the splenic parenchyma (arrowhead) related to the patients leukemia. A calcified splenic artery was also noted (small arrows). TECHNIQUE: mAs 205, kVp 100, slice thickness 4mm.

**Figure 2:** A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: Celiac angiogram. (a) Early arterial image showing moderately dilated gastroduodenal (small arrow) and right gastroepiploic arteries. The splenic artery (large arrow) appears very tortuous. (b) Delayed image shows the right gastroepiploic artery (arrowheads) communicating with the distal splenic artery (small arrow) through anastomosing with the left gastroepiploic artery at the greater curvature of the stomach. TECHNIQUE: 5 French Simmons 2 catheter, (Allura Xper FD20 Systems; Philips, Amsterdam, Netherlands).
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Figure 3: A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: 3D-Volume rendered image demonstrating the anastomosis between the right and left gastroepiploic arteries appears patent (arrowheads), making catheterization of the distal splenic artery from the gastroduodenal artery through the gastroepiploic arteries feasible. TECHNIQUE: 3-D volume rendered image from CTA obtained in the arterial phase. mA: 485. kvp: 120. Slice thickness: 1.25 mm. Contrast agents: 140 mL of Isovue 370 IV contrast.

Figure 4: A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: The microcatheter was advanced through the right gastroepiploic artery and into the left gastroepiploic artery and eventually into the distal splenic artery. Angiogram through the microcatheter confirmed location in the distal splenic artery (arrow). TECHNIQUE: 5 French Simmons 2 catheter, 2.8 French, 150 cm microcatheter, (Allura Xper FD20 Systems; Philips, Amsterdam, Netherlands).

Figure 5: A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: Follow up angiogram through the microcatheter following partial embolization of the lower two-thirds of the spleen using 500 um polyvinyl alcohol particles, shows residual flow in the non-embolized segment of the spleen (arrow). TECHNIQUE: 5 French Simmons 2 catheter, 2.8 French, 150 cm microcatheter, (Allura Xper FD20 Systems; Philips, Amsterdam, Netherlands).

Figure 6: A 77 year old patient with chronic lymphocytic leukemia underwent splenic embolization through gastric collateral circulation. FINDING: Follow up CT scan, coronal reformatted image done 2 years after the procedure revealed marked decrease in the size of the spleen (14.6 cm) with multiple hypodense areas in its lower two-third (arrow) consistent with prior embolization. TECHNIQUE: mAs 201, kVp 100, slice thickness 4mm.
Several causes can contribute to splenomegaly and hypersplenism. Causes can be sub-grouped into: Infectious causes, hyperplastic splenomegaly (like our reported case), congestive splenomegaly, infiltrative splenomegaly and miscellaneous causes.

Almost half of the chronic lymphocytic leukemia patients develop splenomegaly.

More than 50% of the patients with chronic lymphocytic leukemia are over 70 years of age at the time of diagnosis.

Treatment to certain chemicals, Family history, Gender (Male), Race/ethnicity (White race more than Asian race).

Splenectomy, Partial splenic embolization, ligation and banding of the splenic artery, and percutaneous placement of a narrowed stent into the splenic artery.

Splenomegaly is defined as increase in the cranio-caudal length of the spleen more than 11 cm or increase in spleen weight more than 400-500 gm. However, Hypersplenism is only defined based on laboratory results along with bone marrow biopsy.

Table 1: Summary table for splenomegaly and hypersplenism.

<table>
<thead>
<tr>
<th>Causes of splenomegaly and hypersplenism</th>
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<tbody>
<tr>
<td><strong>Infectious causes</strong></td>
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<tr>
<td>- Viral: <em>e.g.</em> infectious mononucleosis.</td>
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<td>- Bacterial: <em>e.g.</em> tuberculosis.</td>
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<td>- Fungal: <em>e.g.</em> histoplasmosis</td>
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<td>- Parasitic: <em>e.g.</em> schistosomiasis</td>
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<tr>
<td><strong>Hyperplastic splenomegaly</strong></td>
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<tr>
<td>- Hereditary spherocytosis.</td>
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<td>- Thalassemia.</td>
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<td>- Polycythaemia rubra vera.</td>
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<td>- Myelofibrosis.</td>
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<td>- Chronic myeloid leukaemia.</td>
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<td>- Chronic lymphocytic leukaemia.</td>
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<tr>
<td>- Lymphoma.</td>
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<tr>
<td><strong>Congestive splenomegaly</strong></td>
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<td>- Liver cirrhosis.</td>
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<td>- Hepatic vein obstruction.</td>
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<td>- Portal vein obstruction.</td>
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<td>- Congestive heart failure with increased venous pressure.</td>
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<tr>
<td><strong>Metabolic and infiltrative splenomegaly</strong></td>
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<tr>
<td>- Gaucher's disease.</td>
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<td>- Amyloidosis.</td>
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<td>- Niemann-Pick disease.</td>
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<td>- Histiocytosis.</td>
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<td>- Marble bone disease.</td>
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<td><strong>Miscellaneous causes</strong></td>
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<td>- Iron deficiency anemia.</td>
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<td>- Vitamin B12 deficiency.</td>
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Table 2: Differential diagnosis table of splenomegaly and hypersplenism.
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<tr>
<th>Imaging modality</th>
<th>General imaging features</th>
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</table>
| Ultrasonography          | - Splenic parenchyma is homogeneous in echogenicity. The spleen can be slightly more echogenic than healthy liver and markedly hyperechoic compared to kidneys.  
- The shape and orientation of the spleen make accurate linear measurement difficult. A length of 11 cm is considered the upper limit of normal size. |
| Computed tomography      | - On non-contrast-enhanced CT images, the healthy spleen usually has a density of around 45 Hounsfield units.  
- The spleen can be considered enlarged if its craniocaudal length is more than 11 cm.  
- A spleen that extends below the lower third pole of the kidney is also indicative of splenomegaly. |
| Magnetic resonance imaging | - On T1-weighted images, the normal signal intensity of the spleen is lower than that of the liver. Conversely, the spleen shows higher signal intensity on T2-weighted images when compared to the liver.  
- Normal craniocaudal length of the spleen should not exceed 11 cm. |

**Table 3:** Imaging of splenomegaly and hypersplenism.

**ABBREVIATIONS**

- CLL = Chronic lymphocytic leukemia  
- CT = Computed tomography  
- PCA = Patient controlled analgesia  
- PSE = Partial splenic embolization  
- VAS = Visual analogue scale

**KEYWORDS**

- Chronic lymphocytic leukemia; Hypersplenism; Splenic artery calcification; Gastro-epiploic collaterals; Splenic embolization