# CASE REPORT

# Adrenal Hemorrhage as a Complication of Plug-assisted Retrograde Transvenous Obliteration of Gastrorenal Shunt Managed by Adrenal Artery Embolization: A Case Report

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#### **Abstract:**

We describe a patient who underwent plug-assisted retrograde transvenous obliteration for gastric varices. After the procedure, the patient developed hypotension and tachycardia. Contrast-enhanced computed tomography showed a left adrenal hematoma. The patient was managed with left inferior adrenal artery embolization. We herein describe an unexpected complication during plug-assisted retrograde transvenous obliteration and the endovascular management by adrenal artery embolization. We speculate that inadvertent cannulation of an adrenal vein tributary and iatrogenic trauma thereafter caused by sheath advancement was a probable cause for this complication. Further increase in intra-adrenal pressure due to blockage of the adrenal vein outflow postplug deployment possibly led to the rupture of adrenal vein tributary and adrenal gland hematoma in our case.

#### **Keywords:**

plug-assisted retrograde transvenous obliteration (PARTO), adrenal hemorrhage, adrenal artery embolization, chronic liver disease (CLD), portal hypertension

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# Introduction

Endovascular treatment of isolated gastric varices involves transvenous retrograde obliteration of the draining gastrorenal shunt by balloon or vascular plug to embolize the varices [1-5]. Adrenal hemorrhage as a complication of shunt obliteration is uncommon. Since adrenal hemorrhages are rare, managing such an iatrogenic adrenal hemorrhage is a challenge [6-8]. We describe an extremely unusual and unanticipated clinical scenario wherein PARTO-induced adrenal hemorrhage was successfully managed by emergency adrenal artery embolization.

## Case Report

Written informed consent for publication of information such as photographs and images has been obtained from the patient. A 54-year-old female patient of Child Pugh A, MELD-12, a known case of hepatitis B-related chronic liver disease, presented with recurrent upper gastrointestinal bleed due to isolated gastric varices for which endoscopic banding was done in the past. CT confirmed insolated gastric varices draining via a solitary gastrorenal shunt. She was planned for PARTO of the shunt.

Using right femoral venous access, the gastrorenal shunt was cannulated (**Fig. 1a**). The 4-Fr Cobra catheter was further advanced into the shunt using the support of a 2.7-Fr Progreat microcatheter (Terumo, Japan). The CFV sheath was exchanged for a 60-cm-long 8 F sheath (Cook, Ind. USA) over a stiff guidewire (Amplatz Boston Scientific, USA). After advancement of the sheath into the shunt, extravasation of contrast was noted likely due to wire and sheath careless manipulation since the wire was likely mistakenly placed in the adrenal vein during sheath advancement. Inadvertent insertion of the sheath dilator into the narrow adrenal vein caused injury to the adrenal gland and ad-

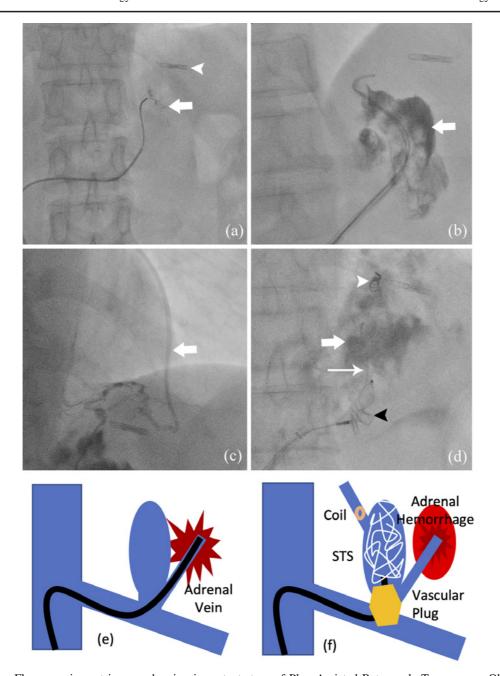
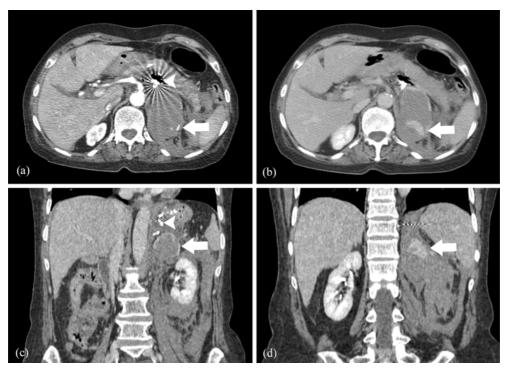


Figure 1. Fluoroscopic spot images showing important steps of Plug Assisted Retrograde Transvenous Obliteration (PARTO) - (a): Using a Cobra catheter, the left renal vein was accessed through femoral vein access. Using the support of the microcatheter, the main Cobra catheter was further advanced. Note the microcatheter inadvertently mistakenly entering and abutting the adrenal vein (white arrow). The previous gastric banding clips also seen (white arrowhead). The illustrated image of this step is shown below in Fig 1. e. (b): The long sheath was advanced over a Stiff Amplatz guidewire. Venography taken after sheath advancement shows contrast extravasation, likely due to adrenal venous ooze secondary to stiff wire manipulation and advancement of sheath through the narrow adrenal vein. (c): The sheath was re-positioned into the gastro-renal shunt. Fluoroscopy spot film also shows a pericardio-phrenic collateral efferent draining channel from the gastric varices (which were embolized with coils); see image Fig 1. d. (d): Embolization of the gastric varices with Sodium Tetradecyl Sulfate (STS): Lipiodol: air foam mixture (white arrow) using a 2.7 Fr Progreat micro-catheter (thin white arrow) advanced through the sheath tracked from the side of the vascular plug (black arrowhead). Note the micro coils (white arrowhead) used to embolize the pericardio-phrenic efferent draining channel shown in the previous image Fig 1. c. The vascular plug is placed at the narrowest portion of the shunt caudal to the entry of the adrenal vein. The corresponding illustration is shown in Fig 1. f. (e): Illustration showing the steps of procedure causing the complication. Mistaken entry of guidewire-sheath into the adrenal vein with careless manipulation causing iatrogenic adrenal vein injury. (f): Illustration showing the mechanism of adrenal haemorrhage. Post PARTO showing vascular plug, Sodium Tetradecyl Sulfate (STS) and micro-coil. Note the position of the vascular plug at the narrowest portion of the gastro-renal shunt caudal to the entry of the adrenal vein. Prior inadvertent adrenal vein damage is responsible for further increased adrenal pressure to outflow obstruction causing adrenal gland damage and resultant adrenal haemorrhage.



**Figure 2.** Post PARTO contrast enhanced Computed Tomography (CECT) - (a): Axial images in the early contrast-enhanced phase show a large adrenal hematoma in left suprarenal location with suspicious active contrast extravasation (white arrow). Note the vascular plug (white arrowhead) with streak artefact, which was deployed during PARTO. (b): Axial scan in the late venous phase shows increase in density of the contrast (white arrow) confirming active haemorrhage. (c): Coronal reformatted image better shows the left adrenal hematoma with suspicious extravasation (white arrow). Note the Lipiodol: Sclerosant in the gastric varices (white arrowhead). (d): Coronal reformatted images confirm the increased density of contrast in the delayed phase confirming active haemorrhage.

renal parenchymal damage, seen as contrast extravasation on the venogram (Fig. 1b). Reinsertion of the wire and sheath into the gastrorenal shunt was then performed. A small draining pericardio-phrenic collateral was noted, which was embolized with microcoil (Fig. 1c). Vascular plug with a diameter of 14 mm (Abott) was deployed at the narrowest portion of the shunt. Complete embolization of the gastric varices was done using a mixture of lipiodol, STS, and air (ratio 1:2:3) (Fig. 1d). The plug was detached and the sheath was removed. The patient complained of progressive abdominal pain and tachycardia 1 h after the procedure. An emergent CT scan (Fig. 2) showed a large left adrenal hematoma. An area of suspicious contrast extravasation was noted, more pronounced in the delayed phase. Since the extravasation was observed in the early contrast-enhanced phase of the contrast CT and that the extravasation was increased in the late contrast-enhanced phase, possibility of adrenal arterial branch as a cause of bleeding was plausible. However, the first suspicion was that of a leak or rupture from the shunt. Femoral vein and resultant left renal vein were cannulated. A contrast venography after placing the catheter abutting the deployed plug showed no contrast extravasation or leak from the left renal vein or shunt (Fig. 3 a). A microwire or microcatheter could not be negotiated through the side of the plug despite repeated attempts. In view of a large adrenal hematoma and vitally unstable patient, an adrenal artery angiography followed by embolization was planned to reduce complete adrenal artery supply. A contrast angiography from the inferior adrenal artery showed contrast extravasation from one of the terminal branches (**Fig. 3b**). There was no contrast extravasation from the superior and middle adrenal arteries on angiography. Superselective cannulation of the branch of the left inferior adrenal artery responsible for extravasation was done followed by glue [glue/lipiodol ratio 1:1] embolization (**Fig. 3c and 3d**). The patient stabilized over the next 6 h and was discharged on day 2 postprocedure.

The patient has shown no fresh symptoms at 6- and 12-week follow-up with no signs of any adrenal insufficiency.

### **Discussion**

Acute adrenal hemorrhage as a severe complication of gastrorenal shunt closure by B-RTO or PARTO is very rare. We postulate that the iatrogenic injury caused during inadvertent sheath advancement into the narrow adrenal vein resulted in adrenal gland damage. The further increase in intra-adrenal venous backpressure due to outflow obstruction after vascular plug deployment at the narrowest part of the gastrorenal shunt ultimately led to adrenal gland damage

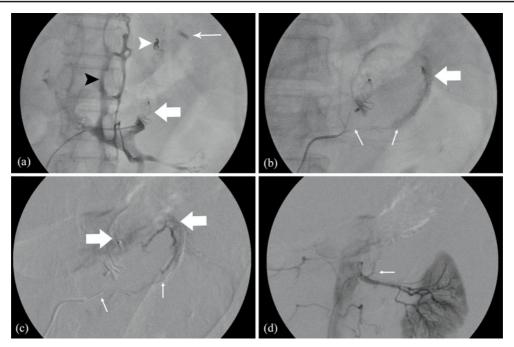


Figure 3. (a): Fluoroscopy spot image shows previously deployed vascular plug (white arrow). A venography taken from a Cobra catheter placed in the gastro-renal shunt at the deployed plug showed no contrast extravasation from the uncovered landing zone of the shunt. Note the filling of the paravertebral venous plexus (black arrowhead). The micro-coils (white arrowhead) deployed during PARTO and the gastric ligation bands (thin white arrow) are also seen. (b): A selective angiogram done via the right femoral arterial access shows active contrast extravasation (thick white arrow) from the left inferior adrenal artery (thin white arrows) arising from the left renal artery. (c): Super selective angiogram after selective cannulation of the left inferior adrenal artery (thin white arrows) using a microcatheter confirms active contrast extravasation (thick white arrows). Superselective cannulation of the branch of the left inferior adrenal artery responsible for extravasation was done followed by glue [Glue:Lipiodol ratio 1:1] embolization. (d): Post embolization angiogram shows a short residual stump of the left inferior adrenal artery (thin white arrow) and no active contrast extravasation. Note the normal left renal artery and renal nephrogram.

and arterial injury [7, 8]. The adrenal vein is a well-known efferent drainage pathway for the gastric varices. The gastric veins in and around the posterosuperior part of the gastric wall anastomose with the inferior phrenic vein (IPV) at the bare area of the stomach. The left IPV terminates inferiorly into the left renal vein often together with the left adrenal vein [9]. The left inferior phrenic vein (LIPV) may have two orifices; one enters the inferior vena cava or the hepatic vein via the subdiaphragmatic transverse part of the LIPV under the left diaphragm, while the other enters the left renal vein (LRV) via the vertical part of the left adrenal vein (LAV) [10]. The latter course has received increased attention because this course may be an origin site for a gastrorenal shunt from gastric varices in patients with portal hypertension. An anastomosis of the LIPV with the gastric or portal vein might occur as a result of the close embryonic development of both organs, which are formed within 4-6 weeks of each other. The venous valve of the LIPV might eventually form the lowest waist portion of the gastrorenal shunt. Vascular plug closure of the shunt caudal to the point where the adrenal vein enters at the narrowest waist of the shunt could theoretically increase the backpressure changes and lead to spontaneous adrenal hemorrhage. Sheath advancement while

the wire was mistakenly parked in the draining adrenal vein is the probable causative triggering contributory factor for initial adrenal bleeding from small adrenal venules in our case. The intraprocedure venous bleed was probably aggravated postprocedure by shunt obliteration, further increasing the intra-adrenal venous pressure (Fig. 4). Min-Yun Chang et al. [3] have postulated a similar theoretical anatomical basis for post-PARTO-induced acute adrenal insufficiency. Such spontaneous adrenal hemorrhages caused by raised adrenal venous pressure can be successfully managed by adrenal artery embolization [6-8]. Various points need to be considered while performing adrenal artery embolization. Care is taken during the procedure to avoid reflux of embolic agents into normal arteries and to avoid contrast extravasation from too forceful an injection. No reports show superiority of one embolic agent over the other. Permanent proximal vessel occlusion can be obtained with microcoils. Gelatin sponges are the least expensive and semipermanent particulate agents include PVA and trisacryl gelatin microspheres can be used in the setting of trauma and for tumor debulking. Liquid embolics can be used in the setting of distal penetration and for small tortuous adrenal vessels, although their usage involves a learning curve and can cause

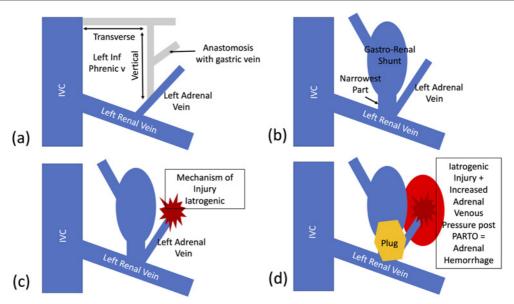


Figure 4. Graphical depiction of plausible etiology of adrenal hemorrhage post Plug Assisted Transvenous Obliteration in our case - (a): Normal anatomy showing the relation of the left adrenal vein (LAV), left inferior phrenic vein (LIPV) and gastrorenal shunt. The left IPV terminates inferiorly into the left renal vein (LRV) often together with the LAV. The LIPV may have two orifices; one enters the inferior vena cava (IVC) or the hepatic vein via the sub-diaphragmatic transverse part of the LIPV under the left diaphragm, while the other enters the LRV via the vertical part of the LAV. The latter course may be an origin site for a gastro-renal shunt from gastric varices in patients with portal hypertension due to close embryological development of both the organs. (b): In portal hypertension, due to increased portal pressures porto-systemic shunts develop at the site where the where LIPV enters LRV via the vertical portion of the LIPV i.e. the gastro-renal shunt with afferent usually from the left gastric vein and efferent into the LRV. (c): In our case, iatrogenic injury from damage to the adrenal vein was noted due to inadvertent mistaken and careless advancement of the sheath into the narrow adrenal vein. (d): Previous iatrogenic adrenal vein trauma and further increased backpressure changes due to vascular plug placement at the narrowest portion of the gastro-renal shunt caudal to the entry of the adrenal vein resulted in adrenal gland trauma, gland rupture and haemorrhage ultimately leading to adrenal arterial injury.

entrapment of the catheter in the embolized artery [11].

Our case report highlights various important features. First, we describe an unreported complication of PARTO and B-RTO. Second, a detailed knowledge of the gastric varices afferent and efferent drainage pathways is essential to better understand the change in hemodynamics post-PARTO and B-RTO. Understanding the anatomical relation between the adrenal vein, gastrorenal shunt, and inferior phrenic vein is important. The vascular sheath should be advanced after the wire has been safely parked in the gastrorenal shunt and not when it is mistakenly placed in the adrenal vein. Placing the vascular plug at the narrowest point of the shunt caudal to the entry of the adrenal vein could potentially further increase adrenal gland venous pressure. Third, it is essential for operators to know that wire and catheter manipulation in the entrance of gastrorenal shunt near the LRV should be very carefully done because the extravasation from the peripheral adrenal vein is not rare. Fourthly, prompt postprocedure monitoring and imaging are essential. Lastly, adrenal artery embolization can be performed in hemodynamically unstable patients for adrenal hemorrhage in the emergency clinical setting. Further studies may be required to observe the short- and long-term changes in adrenal gland function post-PARTO and B-RTO.

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**Author Contribution:** AVK, BAP and KMA performed the study, the procedure and the pre and post procedure patient care; KMA and SP analyzed the data and wrote the manuscript

**IRB:** Ethical approval was waived by the ethics committee due to the retrospective study design.

**Informed Consent:** Consent was unnecessary because no human subject was involved in this study.

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