Case Report

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Isolated Gastric Varix Bleeding Caused by Splenic Vein Obstruction: Two Case Reports

비장 정맥 폐쇄로 인한 단독 위정맥류 출혈: 두 건의 증례 보고

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Isolated left-sided portal hypertension is a rare clinical syndrome caused by splenic veno-occlusive disease. Splenic vein thrombosis and extrinsic compression causes proximal splenic vein hypertension, and the splenic blood flows into the superior mesenteric or portal vein through the upper stomach's collateral vessels, such as the short gastric, coronary, and gastroepiploic veins. Open splenectomy is recommended to treat gastrointestinal bleeding caused by isolated left-sided portal hypertension. Interventional management could be a clinically useful option for selected patients who want to avoid surgical corrections. The report presents two cases of left-sided portal hypertension with gastric variceal bleeding.

Index terms Esophageal and Gastric Varices; Splenic Vein; Hemorrhage; Angioplasty; Therapeutic Embolization

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INTRODUCTION

Gastric variceal bleeding is often associated with liver cirrhosis-related portal hypertension (1). Transjugular intrahepatic portosystemic shunt (TIPS), balloon occluded transvenous retrograde obliteration (BRTO), or percutaneous transhepatic venous obliteration (PTVO) are considered to be treatment options (2). However, inadequate splenic venous drainage can lead to gastric varices and bleeding, although this is rare (3). Here, we report on an attempt to treat two cases of isolated gastric variceal bleeding from splenic vein obstruction with endovascular treatment.

CASE REPORT

CASE 1

A 69-year-old male with alcoholic liver disease and pneumoconiosis visited an outside hospital for hematemesis of three months' duration and underwent endoscopy that revealed gastric variceal bleeding.

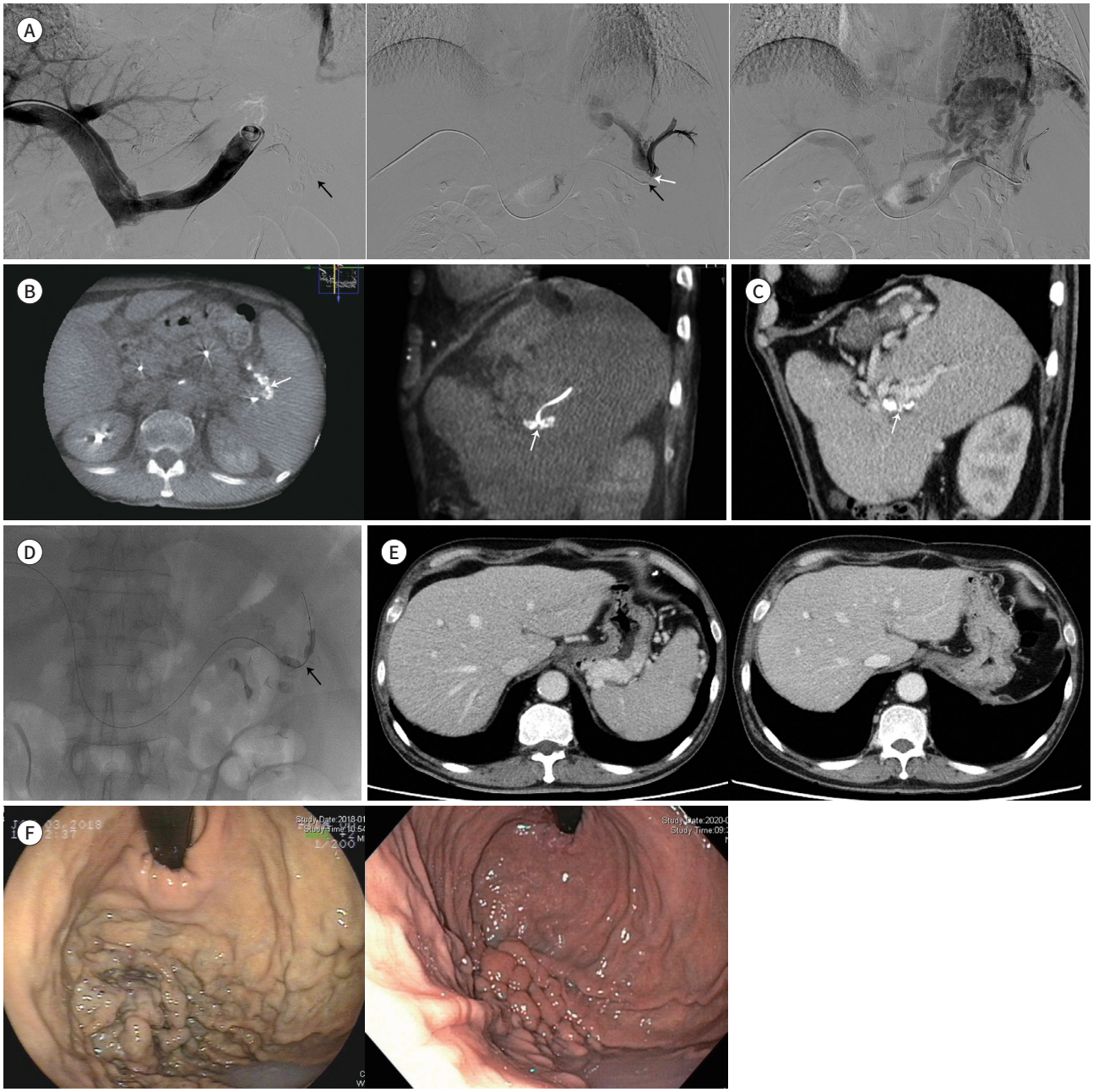
Contrast enhanced abdominal CT images showed gastric fundal varices, which were connected to the splenic vein via the short gastric vein, and which finally drained into the portal vein via the left gastric vein. However, the esophageal varices were indefinite. There was no visible shunt between gastric varices and the systemic vein, such as a gastrorenal or a gastrocaval shunt, or a pericardiophrenic vein. We identified splenomegaly, calcified portocaval, left gastric, and splenic hilar lymph nodes. The findings of liver cirrhosis or abnormal splenic venous drainage were not certain. There was no evidence of pancreatitis. Both lungs showed multiple small nodules on chest radiography which were the sequelae of pneumoconiosis.

Based on CT findings, BRTO was not feasible because of the absence of any shunt between the gastric varices and the systemic vein. Thus, either TIPS or PTVO were available options. After discussion with the referring physician and the patient, we planned to perform PTVO first. In case of a failed PTVO, TIPS would be attempted as a bail-out procedure to reduce portal pressure.

Local anesthesia with 2% lidocaine was performed at the puncture site. The peripheral branch of the segment V portal vein was punctured with a 22-gauge needle (Chiba biopsy needle, Cook Medical, Bloomington, IN, USA) under ultrasound guidance. A 0.018" guidewire (A&A M.D., Seongnam, Korea) was exchanged with a 0.035" guidewire (Roadrunner Uniglide hydrophilic wire, Cook Medical) for the placement of a 5-Fr sheath (Radifocus introducer II, Terumo, Tokyo, Japan). A 5-Fr pigtail catheter (Royal flush plus high-flow catheter, Cook Medical) was advanced toward the distal portion of splenic vein past the junction of the superior mesenteric vein. A splenic venogram with a catheter at the splenic vein demonstrated that the portal blood flow was hepatopetal (Fig. 1A; left). Neither the afferent veins nor the gastric varices were visible. PTVO was not possible because there were no visible afferent veins to access the gastric varix.

Considering CT and angiographic findings, gastric varix was presumed to be caused by isolated left-sided portal hypertension. To demonstrate the splenic venous drainage, a 0.016" -microguidewire (Meister, Asahi, Aichi, Japan) was used, and finally, a 1.98-Fr microcatheter

Fig. 1. Case 1. Isolated gastric varix bleeding caused by splenic vein obstruction. A 69-year-old male with hematemesis.
A. A venogram with a 5-Fr catheter at the proximal splenic vein shows the hepatopetal splenic and portal veins flow but fails to depict the afferent veins and gastric varices. The calcified lymph nodes are at the splenic hilum (arrow) (left image). A venogram with a 1.98-Fr microcatheter placed within the segmental splenic vein shows severe, short-segmental stenosis at the extreme proximal splenic vein from the 5-Fr catheter tip (which cannot pass the stenotic segment) (black arrow) to the contrast-filled venous lumen (white arrow) (middle image). Delayed phase image shows most of the splenic venous drainage divert to the gastric varix via the short gastric vein (right image).
B. Intraoperative cone-beam CT axial (left) and sagittal (right) images. A microcatheter is located between the calcified lymph nodes (arrows).
C. Contrast-enhanced CT sagittal image obtained 4 months preoperatively demonstrates splenic vein stenosis (arrow) between the calcified lymph nodes.
D. Fluoroscopic spot image during percutaneous transluminal angioplasty. The waist of a 6-mm × 40-mm balloon catheter (arrow) suggests short-segmental stenosis at the extreme proximal splenic vein.
E. CT axial images before (left) and after (right) the splenectomy.
F. Endoscopic images before (left) and after (right) the splenectomy. Gastric varices improved on the follow up images.



(Parkway soft, Asahi) was successfully placed within the segmental splenic vein. A splenic venogram showed a severe, short-segmented (1 cm) stenosis at the proximal splenic vein. And most of the splenic venous drainage was diverted to the gastric varix via the short gastric vein (Fig. 1A; middle, right). Cone-beam CT images (Fig. 1B) demonstrated that the stenosis was between the calcified lymph nodes, which were depicted on the patient's abdominal CT images (Fig. 1C). Angiographic findings strongly suggested isolated left-sided portal hypertension caused by severe stenosis at the hilar splenic vein. There were three possible treatment options, including percutaneous transluminal angioplasty (PTA), splenectomy, and partial splenic artery embolization.

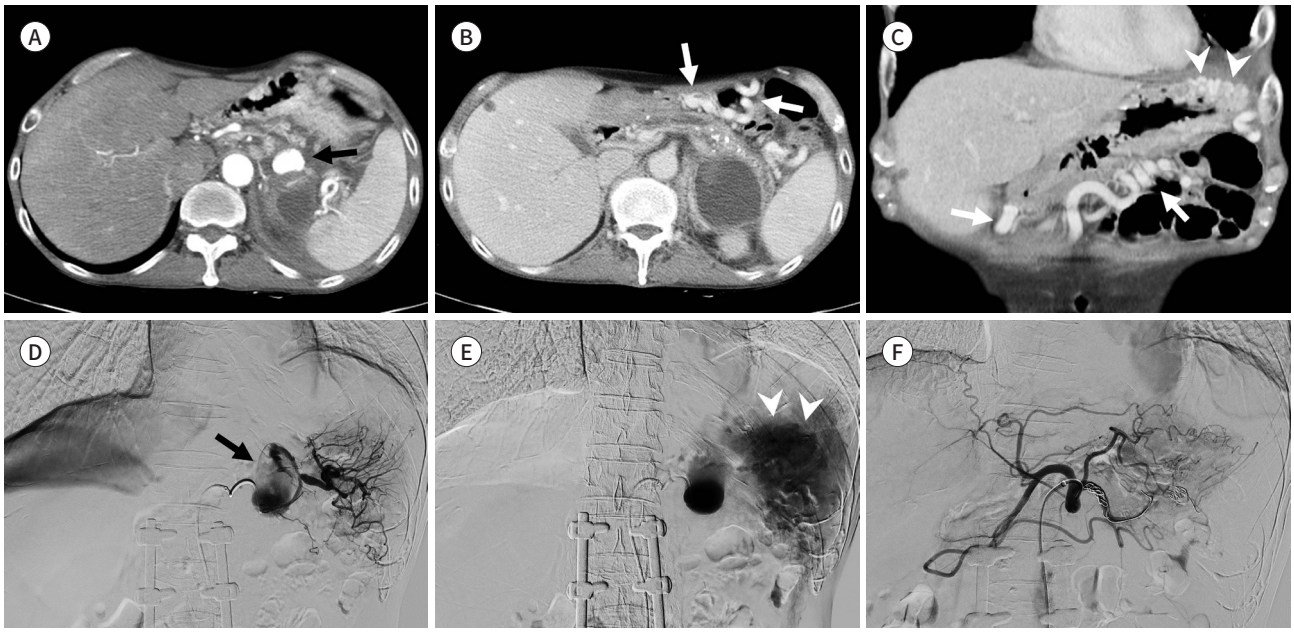
Since the stenotic segment of the splenic vein was short and a microcatheter had been successfully passed through the stenosis, PTA was attempted. After a 0.018"-microguidewire (V-18 control wire, Boston Scientific, Marlborough, MA, USA) had been placed, dilatation was performed with a 4-mm × 40-mm balloon catheter (Savvy long PTA catheter, Cordis, Fremont, CA, USA) and subsequently, with a 6-mm x 40-mm balloon catheter (Saber PTA dilatation catheter, Cordis) (Fig. 1D). Shortly after PTA, his blood pressure, and oxygen saturation started to decrease down to 50/26 mm Hg and 87%, respectively. He became drowsy, with cold sweat. Resuscitation immediately started with intravenous hydration and an oxygen mask. Ultrasonographic examination showed fluid around the spleen, which must have been blood. A splenic venogram with a catheter in the distal splenic vein showed sluggish hepatopetal blood flow. All these findings were suggestive of a splenic vein rupture. The patient successfully underwent emergency splenectomy within three hours following that. Gastric varices improved on follow up CT and endoscopy (Fig. 1E, F). He was free from any bleeding events for 18 months following the operation.

CASE 2

A 57-year-old male visited the emergency room with massive hematemesis (200 cc) and melena started one day ago. Endoscopy revealed the presence of gastric varices, but no active bleeding was found. After 7 days, significant hemoptysis (80 cc) recurred despite conservative management. Vital signs were as follows; blood pressure 80/50 mm Hg, heart rate 120 beats/min,. Laboratory studies revealed elevated serum pancreatic enzyme levels with significant drop of hemoglobin level; amylase 169 U/L, lipase 191 U/L, and hemoglobin 6.3 g/dL. Contrast enhanced abdominal CT images showed atrophic change of pancreas with multiple calcification and pancreatic duct dilatation, suggestive of chronic pancreatitis. A major part of peripancreatic pseudocyst encases the splenic vessels, and concomitant splenic artery pseudoaneurysm (diameter: 1.9 cm) was also noted (Fig. 2). Splenic vein was completely obliterated, and fundal varices with tortuously engorged gastroepiploic veins appear to serve as a collateral venous pathway.

The patient was referred to the radiology department for endovascular management. Since gastric varix was presumed to be caused by splenic venous obstruction on CT findings, TIPS or BRTO seemed to be less effective in this patient. In order to treat both varices of gastroepiploic vein and pseudoaneurysm of splenic artery, transcatheter embolization of splenic artery was planned. For angiographic evaluation, arterial access was obtained via the right common femoral artery using a 5-Fr guiding sheath (Flexor Ansel; Cook Medical). Common hepatic an-

Fig. 2. Case 2. Isolated gastric varx bleeding caused by splenic vein obstruction. A 57-year-old male with hematemesis and melena.
A. The axial arterial phase abdominal CT image reveals a pseudoaneurysm (arrow) arising from the splenic artery.
B, C. The axial and coronal portal venous phase images show fundal varices (arrowheads) and tortuously engorged gastroepiploic veins (arrows). Gross ductal dilatation and extensive calcifications are depicted in the pancreas. The pseudocyst encases splenic vessels, and the splenic vein is not visualized.
D. The arterial-phase image from the splenic angiogram reveals a pseudoaneurysm (arrow) with a wide neck from the splenic artery.
E. The delayed venous-phase image shows fundal varices opacification (arrowheads) without splenic venous enhancement.
F. Immediate post-embolization angiography demonstrates the complete splenic artery and pseudoaneurysm occlusion.



giogram using 5-Fr catheter (Yashiro; Terumo) revealed a huge pseudoaneurysm arising from splenic artery. However, the splenic vein was not visualized until the delayed portal venous phase. A 2.0-Fr microcatheter (Progreat; Terumo) was advanced into the distal circulation of pseudoaneurysm. Distal branches of splenic arteries were embolized with polyvinyl alcohol and Gelfoam particles. Subsequently, successful embolization was achieved with several detachable microcoils (Concerto, Medtronic, Sunnyvale, CA, USA) across the neck of pseudoaneurysm. The further post-procedural course was uneventful and the patient discharged without significant complication 11 days after the procedure. On follow-up, over a period of 6 months, there had been no further episode of variceal bleeding.

This study was approved by the Institutional Review Board of our institution (IRB No. 2022-03-016) and the requirement for informed consent was waived.

DISCUSSION

Isolated left-sided portal hypertension, also called sinistral portal hypertension, is a rare clinical syndrome caused by a splenic vein obstruction (3). Splenic vein thrombosis or extrinsic compression causes hypertension of the proximal splenic vein, and the splenic blood flow drains into the superior mesenteric or portal vein through the collateral vessels in the upper stomach, such as the short gastric, coronary or gastroepiploic veins. This may or may not lead

to gastric varices, according to the anatomical variation, and when the coronary vein carries splenic venous flow, esophageal varices may develop (4). There are various etiologies of splenic vein obstruction, such as pancreatitis, pancreatic neoplasm, pancreatic pseudocyst, gastric, renal, or retroperitoneal pathology, arterial pseudoaneurysm, tuberculous adenitis, or thrombocythemia (4).

Diagnostic modalities of isolated left-sided portal hypertension include barium studies, esophagogastroduodenoscopy, endoscopic US to evaluate gastric varices, transabdominal US, and CT and MR imaging to evaluate splenic and collateral veins and to rule out liver cirrhosis as a cause of portal hypertension. Splenic arteriography is the gold standard for the diagnosis of isolated left-sided portal hypertension, with findings being non-visualization of the segmental or entire splenic vein, despite opacification of splenic parenchyma or portal vein on delayed imaging (5). In our two cases, the gastric varices were prominent on CT. The findings of liver cirrhosis were not defined on CT, but the presence of portal hypertension could not be confirmed because portal pressure was not measured. Multiple calcified lymph nodes around the splenic hilum made it difficult to assess the patency of the splenic vein on CT. However, the short segmental stenosis of the proximal splenic vein was identified on the splenic venogram. Then it was compared to the cone-beam CT taken during the procedure. A splenic vein stenosis between the calcified lymph nodes could be found on CT (Fig. 1F).

Open splenectomy is a recommended treatment for isolated left-sided portal hypertension causing gastrointestinal bleeding (6). Splenectomy in asymptomatic patients with gastric varices remains controversial because of insufficient evidence and insignificant differences in the overall survival rate as compared to conservative management (7). Endovascular treatment has been reported to be another treatment option for variceal bleeding. Splenic arterial embolization may be performed as a pretreatment to splenectomy to reduce blood flow to the spleen (7), and it may be attempted in children who are not adequate for splenectomy or stent insertion. But there is a concern about the long term recurrence of isolated left-sided portal hypertension symptoms (5). Embolization of most of the spleen is not recommended because it can cause more complications (8). Stent insertion for the stenotic splenic vein may be an alternative treatment. There have been reports of stent insertion for significant stenosis of the splenic vein concomitant with a TIPS to reduce portal hypertension (9), and Wallstent insertion for splenic vein stenosis with gastric variceal embolization (10). But, reconstruction of the splenic vein may be performed selectively because these procedures do not have enough data (5).

We have presented two cases of isolated left-sided portal hypertension caused by splenic veno-occlusive disease. Interventional management could be a clinically useful option for selected patients who wish to avoid surgical corrections. However, major complications including vascular rupture may occur and require immediate attention to prevent deleterious consequences.

Author Contributions

Conceptualization, all authors; data curation, all authors; investigation, all authors; methodology, all authors; resources, all authors; supervision, P.J., L.S., L.H.N., H.D., P.S.; validation, P.J., L.H.N.; visualization, L.H.N.; writing—original draft, P.J., L.S.; and writing—review & editing, L.S., L.H.N., H.D., P.S., C.Y.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

1. Ge PS, Runyon BA. Treatment of patients with cirrhosis. *N Engl J Med* 2016;375:767-777
2. Zhang K, Sun X, Wang G, Zhang M, Wu Z, Tian X, et al. Treatment outcomes of percutaneous transhepatic variceal embolization versus transjugular intrahepatic portosystemic shunt for gastric variceal bleeding. *Medicine (Baltimore)* 2019;98:e15464
3. Evans GR, Yellin AE, Weaver FA, Stain SC. Sinistral (left-sided) portal hypertension. *Am Surg* 1990;56:758-763
4. Köklü S, Coban S, Yüksel O, Arhan M. Left-sided portal hypertension. *Dig Dis Sci* 2007;52:1141-1149
5. Cakmak O, Parildar M, Oran I, Sever A, Memis A. Sinistral portal hypertension; imaging findings and endovascular therapy. *Abdom Imaging* 2005;30:208-213
6. Nishiyama T, Iwao N, Myose H, Okamoto T, Fujitomi Y, Chinen M, et al. Splenic vein thrombosis as a consequence of chronic pancreatitis: a study of three cases. *Am J Gastroenterol* 1986;81:1193-1198
7. Sakorafas GH, Sarr MG, Farley DR, Farnell MB. The significance of sinistral portal hypertension complicating chronic pancreatitis. *Am J Surg* 2000;179:129-133
8. Koconis KG, Singh H, Soares G. Partial splenic embolization in the treatment of patients with portal hypertension: a review of the english language literature. *J Vasc Interv Radiol* 2007;18:463-481
9. Cekirge SH, Weiss JP, Foster RG, McLean GK. Stent placement in a splenic vein stenosis after TIPS creation. *Radiology* 1993;189:197-198
10. Stein M, Link DP. Symptomatic spleno-mesenteric-portal venous thrombosis: recanalization and reconstruction with endovascular stents. *J Vasc Interv Radiol* 1999;10:363-371

비장 정맥 폐쇄로 인한 단독 위정맥류 출혈: 두 건의 증례 보고

박주영[1] · 이상준[2*] · 이형남[1] · 현동호[3] · 박성준[4] · 조영종[5]

좌측 단독 문맥고혈압은 비장 정맥이 폐쇄되어 생기는 드문 임상 질환이다. 비장 정맥의 혈전증 혹은 외부 압박으로 인해 근위부 비장정맥의 고혈압이 발생하게 되며, 비장의 혈류는 짧은 위 정맥, 관상정맥, 위대망정맥 같은 측부혈관을 통하여 상장간막 정맥 혹은 문맥으로 흘러가게 된다. 위장관 출혈을 유발하는 좌측 단독 문맥고혈압에는 개복 비장절제술이 첫 번째 치료방법으로 추천된다. 하지만 중재시술을 통한 치료가 수술적 교정을 원치 않는 환자들에게는 임상적으로 유용한 치료 선택지가 될 수 있다. 본문에서는 위정맥류 출혈을 가진 좌측 문맥 고혈압에 대한 두 개의 증례를 다루고자 한다.

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