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Single Case

Hemothorax following Uncomplicated Endoscopic Variceal Sclerotherapy and Ligation for Esophageal Varices

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Keywords

Hemothorax · Sclerotherapy · Ligation · Esophageal varices

Abstract

Endoscopic variceal sclerotherapy and ligation are standard treatment modalities used for the management of esophageal varices. Reportedly, sclerotherapy and ligation are associated with complications such as hematuria, pulmonary thrombus formation, pleural effusion, renal dysfunction, and esophageal stenosis. However, hemothorax following sclerotherapy and ligation has not yet been reported. We treated a patient who presented with liver cirrhosis and polycythemia vera and later developed hemothorax following the above-mentioned procedures. An 86-year-old man diagnosed with liver cirrhosis due to chronic hepatitis type B and alcohol abuse underwent variceal sclerotherapy using ethanolamine oleate to treat his

esophageal varices. Oozing from the esophageal varices continued even after the sclerotherapy procedure; therefore, we performed endoscopic variceal ligation. The patient developed left-sided hemothorax within 24 h after treatment of his varices, and an emergency thoracotomy was performed. A pulmonary ligament of the left lung was bulging and ripping because of mediastinal hematoma, and oozing was noted. Cessation of bleeding was noted after the laceration of the left pulmonary ligament had been sutured. Ours is the first case of hemothorax reported in a patient following an uncomplicated procedure of sclerotherapy and ligation.

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Introduction

Endoscopic variceal sclerotherapy and ligation are common treatment modalities used to control and manage esophageal variceal bleeding [1, 2]. Despite the proven efficacy of both sclerotherapy and ligation for the management of acute variceal bleeding [1, 2], ligation is the first-line therapy because of its safety and ease of use [3]. However, variceal sclerotherapy and ligation are associated with several adverse effects. While chest pain, pulmonary embolism, renal dysfunction, and esophageal stenosis are known to occur as major adverse events associated with sclerotherapy [4–6], ligation could lead to complications such as esophageal laceration, transient dysphagia, chest pain, esophageal stricture, and ulcer-related bleeding [7].

There is growing concern regarding pulmonary complications associated with sclerotherapy, and nonhemorrhagic pleural effusion has been reported after variceal sclerotherapy [8]. A retrospective study reveals that the incidence rate of pleural effusion is about 27% [8]. Of note, hemothorax reportedly occurs in patients following a traumatic accident [9]. Occurrence of massive hemothorax has been reported after blunt trauma leading to injury to the inferior phrenic artery [9]. However, there is not much information regarding the occurrence of hemothorax in association with uncomplicated esophageal variceal sclerotherapy.

We report the occurrence of hemothorax in a patient diagnosed with esophageal varices following an uncomplicated esophageal variceal sclerotherapy and ligation procedure. The patient presented with liver cirrhosis and polycythemia vera with concomitant esophageal varices. After undergoing endoscopic variceal sclerotherapy and ligation, he complained of dull left-sided thoracic pain. Based on the findings of a computed tomography (CT) examination, he was diagnosed as having left-sided hemothorax. Ours is the first report to describe a case where endoscopic variceal sclerotherapy and ligation possibly contributed to the development of hemothorax in a patient.

Case Presentation

An 86-year-old man diagnosed with liver cirrhosis due to chronic hepatitis type B and alcohol abuse was investigated for the presence of esophageal varices at the time of a follow-up visit to the Department of Gastroenterology at our hospital. He had a history of left-sided intramuscular hemorrhage of unknown etiology, a year prior to presentation. He had been

diagnosed with polycythemia vera at the age of 74 years, but he did not relate any remarkable family history. A physical examination revealed he was 150 cm tall and weighed 55 kg. An examination of his palpebral conjunctiva did not reveal an anemic state, and his bulbar conjunctiva did not show signs of icterus. His heart and respiratory sounds were normal, and his liver and spleen were not palpable. A laboratory workup revealed a red blood cell count of $5.36 \times 10^6/\mu\text{L}$, a hemoglobin level of 13.8 g/dL, and a platelet count of $42.8 \times 10^4/\mu\text{L}$, and his prothrombin time was prolonged in 60%. However, his von Willebrand factor (vWF) was normal. The serum aspartate aminotransferase and alanine aminotransferase levels were elevated to 44 and 40 U/L, respectively; however, the serum albumin, total cholesterol, and triglyceride levels were decreased (Table 1). His hepatic reserve showed a Child-Pugh class B.

We performed an upper gastrointestinal endoscopy to assess for any gastrointestinal complications associated with liver cirrhosis. The endoscopy showed erythema and cherry red spots in the lower part of the esophagus (Fig. 1a). On the second day after admission, he underwent endoscopic variceal sclerotherapy with injection of ethanolamine oleate into the variceal veins to prevent bleeding from the esophageal varices (Fig. 1b, c). After the endoscopic variceal sclerotherapy, the patient developed epigastric abdominal pain and reported tarry stool. The following day, we performed an upper gastrointestinal endoscopy to identify the source of the bleeding leading to the tarry stool. A huge hematoma was detected at the puncture site through which the sclerotherapy had been administered, and we performed endoscopic variceal ligation using an O-ring (Fig. 1d, e). A laboratory workup revealed that his red blood cell count had decreased to $4.6 \times 10^6/\mu\text{L}$, his hemoglobin level had dropped to 11.7 g/dL, and his platelet count was significantly increased to $98.8 \times 10^4/\mu\text{L}$, for which he received urgent blood transfusion.

The following day, the patient complained of severe left-sided dull pain, and had difficulty with breathing. An emergency enhanced CT revealed a massive left-sided pleural effusion, which was suspected to be caused by extravasation from vessels present along the left lung ligament (Fig. 2a). Another laboratory workup revealed that his red blood cell count had further decreased to $3.4 \times 10^6/\mu\text{L}$, his hemoglobin level had further dropped to 8.9 g/dL, and the platelet count had risen to $95.1 \times 10^4/\mu\text{L}$.

On the third day of hospitalization, the patient underwent an emergency thoracotomy, which revealed massive bloody pleural effusion and a huge hematoma in the left thoracic cavity (Fig. 2b). A pulmonary ligament was bulging and ripping because of the mediastinal hematoma, and oozing was noted. The responsible vessel for hemothorax was not clearly identified. We sutured the lacerated portion of the left pulmonary ligament. After removal of the huge hematoma, he received blood transfusion, and no further bleeding was observed. A further laboratory workup revealed that his red blood cell count had returned to $3.7 \times 10^6/\mu\text{L}$, his hemoglobin level had returned to 10.7 g/dL, and the platelet count was $31.3 \times 10^4/\mu\text{L}$. The patient was placed in the intensive medical care unit for observation and medical management.

On the eighth day of hospitalization, he was transferred to the general ward. A CT examination showed that his left-sided massive pleural effusion had decreased (Fig. 3a), and endoscopy revealed thrombus formation in the variceal veins in addition to the presence of a post-banding ulcer after use of the elastic O-ring (Fig. 3b). He was discharged from the hospital 10 days after surgery for evacuation of the mediastinal hematoma.

Discussion

Pulmonary embolism, renal dysfunction, and esophageal stenosis are known to be major adverse effects associated with sclerotherapy [4–6]. Variceal ligation is associated with complications such as esophageal laceration, transient dysphagia, chest pain, esophageal stricture, and ulcer-related bleeding [7]. However, hemothorax following uncomplicated esophageal variceal sclerotherapy and ligation has not yet been reported.

Life-threatening hemothorax has been reported due to injury to the inferior pulmonary ligament after trauma [9]. A patient hit by a car is known to have developed active extravasation of the contrast medium [9]. Massive hemothorax has been reported due to inferior phrenic artery injury after blunt trauma. However, in our case, there was no trauma reported during sclerotherapy and ligation, nor was there any injury to the pulmonary ligament.

To date, only 1 case of hemothorax has been reported following a sclerotherapy procedure performed for esophageal varices [10]. The patient is known to have developed left-sided bloody pleural effusion within 12–72 h after sclerotherapy. The site of bleeding that led to the hemothorax following esophageal variceal sclerotherapy remains unclear. It was hypothesized that hemothorax reflects the severity of inflammation after paravariceal extravasation of the sclerosant. Alternatively, the patient could have had abnormally dilated vessels on the outer wall of the esophagus secondary to portal hypertension. However, in our case, paravariceal extravasation of the sclerosant was not detected. Sclerotherapy itself might induce portal hypertension associated with thrombosis in the treated veins. Changes in hemodynamic status might be a mechanism that contributes to the development of hemothorax after variceal sclerotherapy and ligation.

Polycythemia vera is associated with bleeding primarily involving the skin and mucous membranes, suggesting defective primary hemostasis [11]. Although gastrointestinal hemorrhage occurs less frequently, it could be severe, and is often associated with use of aspirin [12, 13]. This type of a bleeding pattern is consistent with qualitative or quantitative defects in platelets or the presence of von Willebrand disease. Some episodes of hemorrhage may be directly or indirectly related to concomitant thrombotic complications. Previous reports have indicated that bleeding gastric and esophageal varices usually result from portal hypertension associated with thrombosis of abdominal veins [14]. In the present case, the patient had related a history of left-sided intramuscular hemorrhage. His platelet count had increased after esophageal variceal sclerotherapy and rose further after esophageal variceal ligation. Furthermore, he showed a drop in his platelet count with no further episodes of bleeding observed following surgery for treatment of hemothorax and hematoma evacuation. Previous reports have indicated that an elevated platelet count may be associated with an abnormal vWF multimer distribution in plasma [15]. An elevated platelet count did show a correlation with a decrease in the largest multimers of plasma vWF [15]. An inverse correlation is known to exist between the proportion of large vWF multimers and platelets. These findings indicate that an increased platelet count following sclerotherapy might induce qualitative defects in platelets, leading to a greater tendency towards bleeding.

In conclusion, we described a case of hemothorax in a patient following uncomplicated endoscopic variceal sclerotherapy and ligation for the management of esophageal varices. The uncomplicated hemothorax could be attributed to portal hypertension caused by sclero-

therapy-induced vein thrombosis and qualitative platelet defects that led to accelerated bleeding.

Statement of Ethics

Consent was obtained from the patient for the publication of this case report.

Disclosure Statement

All authors declare no conflicts of interest.

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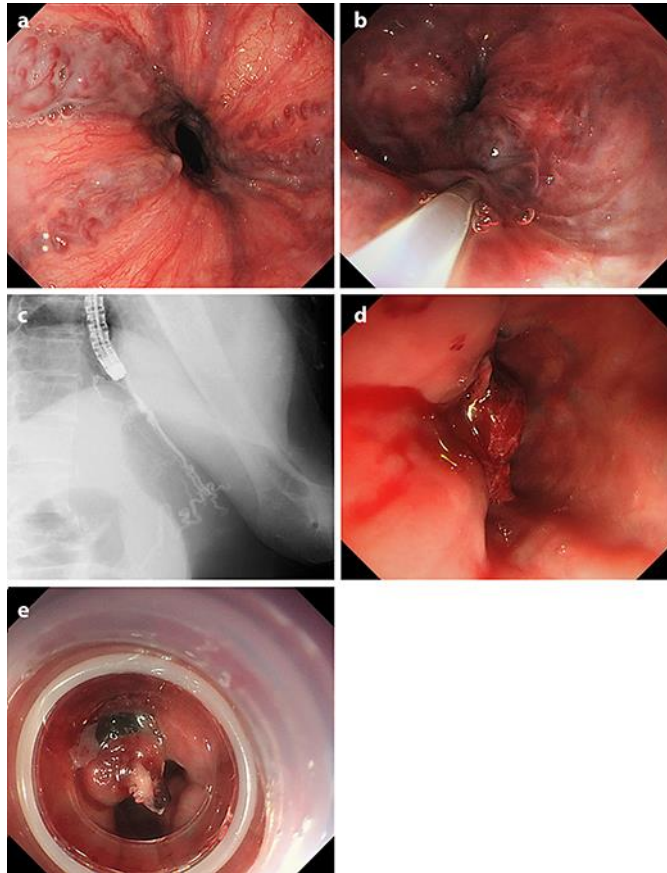


Fig. 1. Endoscopic findings showing tense and nodular varices with cherry red spots at the locus inferior (a). Endoscopic variceal sclerotherapy was performed via intravariceal injection of 5% ethanolamine oleate using a 25-G needle injector (b). Endoscopic varicelography shows the 5% ethanolamine oleate injected into the veins (c). After an endoscopic sclerotherapy procedure performed for the esophageal varices, the endoscopic findings show a blood clot covering the ruptured esophageal varices (d). e Endoscopic variceal ligation.

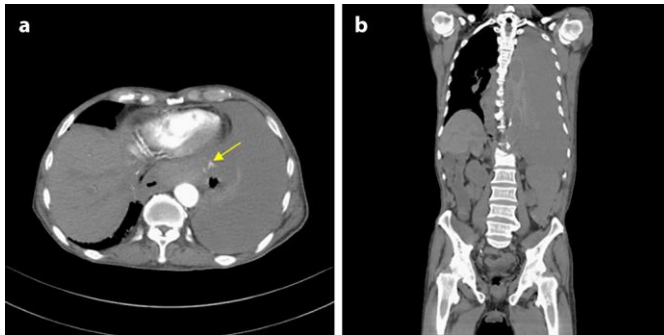


Fig. 2. **a** Coronal view of the patient's chest region noted on a computed tomography (CT) scan, demonstrating significant hemothorax with extravasation of the contrast material presumably from the vessels along the pulmonary ligament. An arrow indicates the vessels along the pulmonary ligament. **b** Axial view of the patient's chest on a CT scan showing the massive hemothorax.

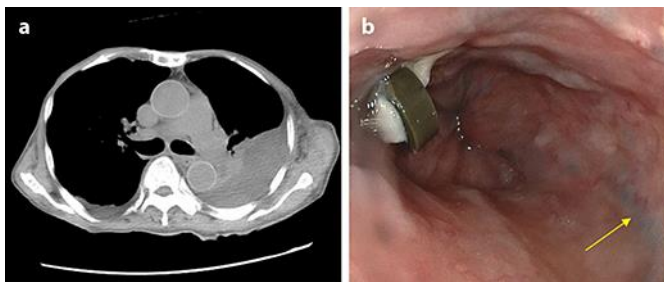


Fig. 3. **a** Coronal view of the patient's chest on a CT scan showing the reduction in the left-sided pleural effusion. **b** Endoscopic finding showing a post-banding ulcer and thrombus formation in the variceal vein. An arrow indicates the vessel with thrombus formation.

Table 1. Laboratory data on the patient at admission

Laboratory data			Hemogram		
Total protein	8.5	g/dL	WBC	39,500	/ μ L
Albumin	3.5	g/dL	RBC	5.36×10^6	/ μ L
Total bilirubin	0.74	g/dL	Hb	13.8	g/dL
BUN	48.8	mg/dL	Ht	45.8	%
Creatinine	1.18	mg/dL	plt	42.8×10^4	/ μ L
AST	44	IU/L	PT%	60	%
ALT	40	IU/L	PIC	0.6	μ g/mL
ALP	513	IU/L	TAT	2.9	ng/mL
LDH	267	IU/L	vWF	56	%
γ -GTP	206	IU/L	Factor XIII	67	%
Glucose	97	mg/dL	AFP	0.8	ng/dL
Total cholesterol	114	mg/dL	PIVKA-II	13	mAU/mL
LDL cholesterol	57	mg/dL	HBsAg	(+)	
Triglyceride	45	mg/dL	HBeAg	(-)	
Fe	17	μ g/dL	HBeAb	(+)	
Ferritin	38.4	μ g/L	HCVAb	(-)	

BUN, blood urea nitrogen; AST, aspartate aminotransferase; ALT, alanine aminotransferase; LDH, lactate dehydrogenase; γ -GTP, γ -glutamyl transpeptidase; LDL, low-density lipoprotein; WBC, white blood cell count; RBC, red blood cell count; plt, platelets; PT%, prothrombin time in percent; PIC, plasmin- α_2 plasmin inhibitor complex; TAT, thrombin-antithrombin complex; vWF, von Willebrand factor; AFP, α -fetoprotein; PIVKA-II, prothrombin induced by vitamin K absence or antagonist-II; HBsAg, hepatitis B surface antigen; HBeAg, hepatitis B e antigen; HBeAb, hepatitis B e antibody; HCVAb, hepatitis C virus antibody.