Recurrence vallecular varices: a rare cause of hemorrhage from the upper aerodigestive tract

Patricia McAdams, MD; Anne Gunter, BS; Del Sloneker, MD; Wayne Harsha, MD
Department of Otolaryngology, Madigan Army Medical Center

ABSTRACT

Objectives: To raise awareness of a rare and diagnostically challenging disease entity that has the potential to recur.

Study design: Case report and literature review.

Methods: Presentation of the only known case of recurrent vallecular varices requiring multiple surgical interventions.

Results: Patients with hemorrhaging vallecular varices are frequently misdiagnosed, as bleeding is often attributed to a gastrointestinal or pulmonary source. We herein describe the only reported case of recurrent varices after surgical management.

A 56 yo male with a history of hepatitis B/C co-infection and cirrhosis presented to the emergency department hemorrhaging from the mouth. While upper GI endoscopy revealed only blood above the esophageal inlet, flexible laryngoscopy demonstrated active bleeding from a prominent vallecular varix. The patient underwent diagnostic laryngoscopy with cautery; however, five months later, the patient presented again with similar symptoms, and multiple recurrent varices were identified on examination. The patient required repeat cautery and has had no further recurrences 9 months postoperatively. Six previous case reports of bleeding vallecular varices were reviewed, and all cases were managed definitively with a single procedure.

Conclusions: The etiology of vallecular varices remains unidentified, although most cases are associated with COPD and/or cirrhosis of the liver. Vallecular venous anastomoses between the portal and systemic systems may be present in affected individuals. Due to the risk of recurrence after operative management, routine postoperative surveillance may prevent future episodes of bleeding.

INTRODUCTION

In 1967, Wetherill and Gandhi detailed one of the first reported cases of spontaneous hemorrhage from a suprathyroidal source, an enlarged vein at the base of the tongue (1). In their discussion, they encouraged examination of the base of the tongue, hypopharynx, and larynx in patients presenting with hemoptysis if no other source of bleeding is found.

Vallecular varices are a rare, yet potentially life-threatening source of bleeding, with only 6 cases in the English literature reported since 1967 (1-6). Patients with vallecular varices can present with hemoptysis, hematemia, or both; these symptoms often compel a physician to suspect cardiopulmonary disease or upper gastrointestinal (GI) tract pathology as the likely etiology. However, preoccupation with identifying a lesion within these organ systems may postpone definitive treatment, as the region of the hypopharynx is easily overlooked by endoscopy and bronchoscopy alike.

Complications due to hemodynamic instability may occur in severe cases of vallecular hemorrhage. This case report aims to emphasize the necessity of timely diagnosis and offer treatment recommendations for patients with variceal lesions discovered in this area. We discuss a patient with a past history of esophageal varices who presented with expectoration of frank blood in the setting of a normal upper GI endoscopy. He was subsequently found to have actively bleeding vallecular varices that proved refractory to electrocautery.

CASE REPORT

The patient is a 56 year old male with a 10 year history of hepatitis B/C co-infection and cirrhosis, who presented to the emergency department with expectoration of blood from the mouth seven times a day for one week. Hematocrit was 39 on presentation. Upper GI endoscopy did not reveal any suspicious lesions; however, blood was noted above the esophageal inlet. Otolaryngology was consulted and performed flexible laryngoscopy, which revealed pooling of blood in the right pyriform sinus and bleeding vallecular varices. The patient underwent diagnostic laryngoscopy with cautery.

The patient had an additional episode of hemorrhage 2 weeks postoperatively. A clot was removed from the surgical site and this area was cauterized. No varices were noted at this time.

Five months later, the patient expectorated blood for a third time. Recurrent varices were noted on exam. He underwent direct laryngoscopy (Figure 1 & 2) and cautery (Figure 3). Nine months after the last intervention, there have been no further reported episodes of hemorrhage, and the varicale appeared normal on exam (Figure 4).

DISCUSSION

Spontaneous bleeding from the oropharynx is often attributed to a GI, pulmonary or nasal source. Patients with hemorrhage from vallecular varices frequently undergo extensive work-up prior to definitive diagnosis.

A literature review yielded 9 prior cases of hemorrhage from variceal lesions of the tongue base or vallecula. Four patients had COPD (1-4) and one patient had cirrhosis with portal hypertension similar to our patient (5). The fact that these comorbid conditions are commonly associated with hemorrhage makes diagnosis even more difficult, as the bleeding is often missed for hematemesis or hemoptysis. Four patients underwent both bronchoscopy and upper GI endoscopy prior to diagnosis of variceal lesions on the base of tongue or vallecula, and some of these patients had multiple admissions with repeat diagnostic procedures.

When the source of bleeding is not readily identified, the clinician should consider unusual conditions and anatomic regions such as the vallecula. Delayed diagnosis may result in significant morbidity. Three patients required blood transfusions (2,4,6). One patient underwent an exploratory laparotomy for a presumed massive GI hemorrhage (2), and another required tracheostomy (4). Definitive treatment of these lesions has been described with cauteterization, carbon dioxide laser treatment, and sclerotherapy.

Our patient underwent a total of 3 cauteterization procedures. The hemorrhage preceding the second procedure was attributed to sloughing of the eschar, and return of the variceal lesions. However, five months later, the patient experienced hemorrhage from recurrent variceal lesions which were definitively cauterized. No vallecular varices were present on exam 9 months later.

CONCLUSION

Vallecular varices are a rare cause of hemorrhage that may result in misdiagnosis and significant morbidity.

While the etiology of vallecular varices is unknown, most cases are associated with COPD. Elevated right heart pressures may cause distension of veins in the vallecula, resulting in varices. One previous case was associated with chronic sinus similar to our patient. A rare anatomic variant with anastomoses between the portal and systemic venous systems in the vallecula may explain this presentation.

Our experience suggests there may be a role for routine postoperative surveillance in these patients to monitor for recurrence.

REFERENCES