

Base of Tongue Varices; A Rare Cause of Significant Hematemesis in Cirrhotic Patient with Portal Hypertension: A Case Report

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Abstract

Patients with advanced liver disease can develop life-threatening bleeding and have increased bleeding risk due to portal hypertension and coagulopathy. Bleeding from lingual varices is a rare but important source which can be readily missed if not considered. The relationship of lingual varices with portal hypertension is not clear. This case report highlights lateral thinking on causes of hematemesis and careful examination.

Keywords: Tongue Varices; Hematemesis; Cirrhotic Patient; Portal Hypertension

Introduction

Patients with advanced liver disease can develop life-threatening bleeding and have increased bleeding risk due to portal hypertension and coagulopathy.

Case Report

A 55 year old gentleman with a history of cirrhosis and portal hypertension secondary to alcohol-related liver disease presented with hematemesis with no significant drop in haemoglobin (101 g/L) and normal urea (4.8 mmol/L). An emergency oesophago-gastro-duodenoscopy (OGD) showed no evidence of active or recent bleeding, and no oesophageal or gastric varices. Haemoglobin and other parameters remained stable, and the patient was discharged. Two weeks later, he re-presented to the Emergency Department with witnessed haematemesis of approximately 500 ml, associated with a drop in haemoglobin to 87 g/L, but with normal urea (4.1 mmol/L). Once again, OGD failed to show a bleeding source. Given the rapid recurrence, absence of melaena, and normal urea an urgent CT thorax with contrast was arranged to exclude the possibility of haemoptysis secondary to thoracic pathology. The images confirmed decompensated chronic liver disease but no pulmonary embolism, mediastinal hematoma or broncho-arterial fistula. The patient continued to have recurrent fresh hematemesis with associated fall in haemoglobin (83 g/L), but without hemodynamic compromise or urea rise. A CT angiogram from base of skull to pelvis did not identify any bleeding point in the upper aero-digestive tract, therefore the medical team considered the possibility of bleeding from an atypical source.

The patient was reviewed by otorhinolaryngology [ENT], who performed a posterior laryngoscopy, which was unremarkable. Shortly afterwards the patient was witnessed to have spontaneous blood pooling in the mouth without associated coughing or vomiting. On

examination of the oral cavity, anterolateral tongue bruising was observed in close proximity of an engorged lingual vein. He was subsequently referred to the maxillofacial surgeons, who confirmed the presence of dilated lingual varicosities. Following cauterisation, there was no further bleeding. The patient was discharged after 2 days and has reported no further bleeding.

Patient and team perspective

We asked the patient and his wife how his experience affected them. He described his first emotion as fear. Both he and his wife were very concerned, especially as the problem was recurrent and without explanation. They were afraid he would be discharged again without a diagnosis, and he feared for his life. When the source of bleeding was identified, the patient told us that he had a sense of relief and was very grateful to the surgeon who treated him.

This case was also an important learning event for the medical team, involving as it did a highly unusual cause of recurrent bleeding, demonstrating the importance of thorough clinical examination, and consideration of alternative aetiologies.

Discussion

Bleeding from the upper gastrointestinal tract most commonly occurs due to peptic ulcer disease, Mallory-weiss tears, oesophagitis or rupture of varices, and can cause significant morbidity and mortality [1].

Suspicion of variceal bleeding secondary to portal hypertension should be high in patients with advanced liver disease. Portal hypertension is a late and highly clinically significant complication of cirrhosis where the hepatic venous pressure gradient exceeds 5 mmHg. The resultant high pressure in the portal system results in porto-systemic shunting and development of collaterals, most commonly in the oesophagus, rectum, para umbilical veins and veins of Retzius [5]. The risk of variceal bleeding increases with rising portal pressure, but can occur at any stage once hepatic venous pressure gradient exceeds 10 - 12 mmHg.

Lingual varices are spontaneous entities which are found in up to 60% of the elderly population [2]. Anatomically, dorsal lingual veins join to form the lingual vein, which runs along the lingual artery on the ventral side of the tongue. It empties into internal jugular vein at the level of the hyoid bone. Lingual varices have been associated with ageing, smoking, cardiovascular disease and hypertension [2] but there is no evidence in the literature that sublingual veins, in isolation, cause potentially life-threatening bleeding. Importantly, there is no previously recognised communication between lingual venous drainage and the portal circulation [3], although a case of dorsal tongue base varices causing significant bleeding in a patient with portal hypertension has been reported previously [4]. In the case described, we consider it most likely that the presence of lingual varices was not directly related to portal hypertension, but that the resultant hyperdynamic circulation and higher bleeding risk from coagulopathy led to more clinically significant bleeding than would otherwise have occurred. It is also possible, but in our opinion far less likely that the patient may have had an atypical and previously unrecognised porto-systemic shunt directly affecting the lingual vein.

Conclusion

Lingual varices can be a rare cause of significant bleeding from upper aero-digestive tract in patients with advanced liver disease and should be considered in cases of recurrent bleeding with no identifiable source on OGD. The case emphasises the need for a thorough clinical examination at every encounter even when a diagnosis seems obvious.

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