CASE REPORT

Where's the ulcer? Spontaneous bleeding from Zenker's diverticulum

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SUMMARY

An 89-year-old man with a history of coronary artery disease status post coronary artery bypass grafting and atrial fibrillation on aspirin and warfarin, presented to the emergency department with a symptom of haemoptysis. He reported a history of dysphagia for 3 months prior to presentation, but had yet to seek medical attention. On presentation, he was hypotensive and tachycardic with a haemoglobin of 6.6 g/dL and an International Normalised Ratio (INR) of 3.9. Esophagogastroduodenoscopy was performed and showed active bleeding from Zenker's diverticulum. The patient's INR was reversed with fresh frozen plasma and his anaemia was treated with packed red blood cells. The bleeding stopped after reversal of his INR. A barium swallow confirmed a 9 cm Zenker's diverticulum. The patient had a diverticulectomy and cricopharyngeal myotomy. Histology showed mild chronic inflammation of the mucosa, but no ulceration. The patient was discharged without any further bleeding episodes.

BACKGROUND

Bleeding from Zenker's diverticulum has been reported in several cases with a common underlying denominator: the presence of an ulcer secondary to aspirin or non-steroidal anti-inflammatory drug (NSAID) use. To the best of our knowledge, there is only one reported case of suspected bleeding from Zenker's diverticulum that was not associated with an ulcer. We present a case of a patient who developed significant gastrointestinal bleeding from Zenker's diverticulum. While our patient was also on aspirin, there was no evidence of ulceration noted during the endoscopy or histology. This case illustrates a spontaneous bleeding from Zenker's diverticulum with no associated ulceration.

CASE PRESENTATION

An 89-year-old man with a medical history significant for coronary artery disease status post coronary artery bypass grafting and atrial fibrillation for which he was on aspirin and warfarin, respectively, presented to the emergency department with a symptom of haemoptysis. He had no prior episodes of overt bleeding. Symptoms began acutely the night prior to admission. He did not report persistent cough, but rather only coughed when he felt that blood had collected at the back of his throat. He denied any nausea, vomiting, abdominal pain, haematochezia or melena. He reported of intermittent dysphagia to solids and liquids over the past 3 months, although he had yet to

seek medical attention. He had never had an esophagogastroduodenoscopy.

INVESTIGATIONS

On examination, the patient was hypotensive, tachycardic and had dried blood in his posterior pharynx. Laboratory data shortly after presentation were significant for haemoglobin of 6.6 g/dL, International Normalised Ratio (INR) of 3.9, blood urea nitrogen (BUN) of 48 and creatinine 1.48 mg/dL. A CT of his chest showed a dilated proximal thoracic oesophagus containing endoluminal gas and debris with focal narrowing distal to this level

Figure 1 (A and B) Active bleeding from Zenker's diverticulum.

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concerning for an oesophageal stricture. Laryngoscopy was performed, but was unrevealing for the source of the bleeding. A nasogastric tube was placed from which there was bright red blood return. The patient was transfused with four units of packed red blood cells, four units of fresh frozen plasma and was intubated for airway protection prior to endoscopy. Upper endoscopy was performed and showed a diverticulum that was actively bleeding immediately distal to the cricopharyngeus (figure 1A, B). No lesion could be identified within the diverticulum due to profuse bleeding, thus no intervention was performed endoscopically. The remainder of the endoscopic examination was normal. There was no stricture noted endoscopically. A barium swallow was performed and confirmed the 9 cm Zenker's diverticulum in the proximal oesophagus as well as a 2 cm mid-thoracic oesophageal diverticulum.

TREATMENT

After the procedure, the patient was transferred to the intensive care unit (ICU) where he continued to receive fresh frozen plasma and vitamin K in an effort to correct his INR. With these measures, his INR improved to 1.5, and he did not have any further bleeding episodes and was extubated. The patient then underwent a diverticulectomy and cricopharyngeal myotomy.

OUTCOME AND FOLLOW-UP

Histology from the surgical specimen showed that there was mild chronic inflammation of the mucosa and associated reactive changes, but no evidence of ulceration (figure 2). The patient was discharged on postoperative day 2 after the resolution of his symptoms on aspirin, but not warfarin.

DISCUSSION

Zenker's diverticula are classified as pulsion diverticula and form due to increased intraluminal pressure leading to an out-pouching of the mucosa in an area of wall weakness.^{1 2} It is difficult to estimate the true incidence of Zenker's diverticulum as many patients are asymptomatic. Bleeding from Zenker's diverticulum, however, has only been described in few case reports, and is thus not a diagnosis routinely suspected in a patient presenting with upper gastrointestinal bleeding.³⁻⁹ The location of Zenker's diverticulum, proximal to the upper oesophageal sphincter, may even lead to a clinical presentation of haemoptysis concerning for a pulmonary source as was the case with our patient.^{6 8}

Figure 2 Diverticulum with mild chronic inflammation, but no ulceration.

Aspirin use was first described as a risk factor for bleeding from Zenker's diverticulum in a patient taking large doses of aspirin.⁴ This association was seen as well in a patient who was taking aspirin only intermittently and in another patient who was taking clopidogrel as well as aspirin.^{6 8} The common finding in these patients was the presence of ulceration seen either endoscopically or on histology after resection. Of the previously reported cases, only one patient developed bleeding from Zenker's diverticulum without evidence of ulceration, although there was no history given of either aspirin or NSAID use.³ Our patient, who did report a history of aspirin use, was not found to have any ulcers on histology. In addition, our patient was also on warfarin therapy.

In the two most recently reported cases of bleeding from Zenker's diverticulum, a visible vessel was seen in the diverticulum and, in both cases, it was successfully clipped without any further episodes of bleeding.^{8 9} Visualisation of the diverticulum was obscured in our patient due to the active bleeding precluding any attempt at endoscopic intervention.

While bleeding from Zenker's diverticulum is a rare occurrence, it can lead to significant morbidity or mortality if not diagnosed and treated appropriately. As their clinical presentation can be misleading, it is important to have a high index of suspicion especially in a patient who presents with atypical symptoms of an upper gastrointestinal bleeding in the setting of recent symptoms of dysphagia. Aspirin use has been associated with ulcer formation in Zenker's diverticulum, but our case illustrates that this pathology is not always required for bleeding to occur. Thus, patients with a known Zenker's diverticulum should have their need for aspirin re-evaluated and, if absolutely necessary, the patients should be counselled on this additional risk of bleeding. If bleeding occurs and a visible vessel can be identified, previous case reports have shown that endoscopic therapy with endoclip placement is a viable therapeutic option.^{8 9} Although endoscopic treatment can be successful, it is often difficult to control the bleeding due to the position of the vessel, especially in the face of acute bleeding, as described in this case. Surgical resection is a definite treatment alternative for Zenker's diverticulum and should be considered if the patient is a good surgical candidate. Once the diagnosis is established, and even if endoscopic therapy is successful at controlling the bleeding, surgery should be contemplated as soon as possible. In cases of active bleeding that fail to respond to endoscopic therapy, emergency surgery should be the treatment of choice.

Learning points

- ▶ Bleeding from Zenker's diverticulum, though rare, should be suspected in an elderly patient who presents with an atypical gastrointestinal bleeding.
- ▶ Aspirin use has been associated with bleeding from Zenker's diverticulum and may lead to bleeding even in the absence of ulcer formation.
- ▶ The patient with a known Zenker's diverticulum should be counselled on the risk of bleeding and the necessity of aspirin should be determined.
- ▶ Endoscopic therapy is a viable therapeutic option in a patient with bleeding from a Zenker's diverticulum if the bleeding source can be identified.
- ▶ Emergency surgery should be the treatment of choice in cases of active bleeding that fail to respond to endoscopic therapy.

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