Aortoduodenalna fistula - usoden zaplet pri bolniku z aortno protezo

Aortoduodenal fistula - fatal complication in a patient with aortic prosthesis

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Kliučne besede:

aorto-enterična fistula, krvavitev iz zgornje prebavne cevi, nujna endoskopija, izkrvavitev.

Kev words:

aorto-enteric fistula, upper gastrointestinal haemorrhage, emergency endoscopy, exsanguination.

Članek prispel / Received 08.07.2009 Članek sprejet / Accepted 11.03.2010

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Izvleček

Namen: Aorto-enterična fistula je izjemno redek vzrok krvavitve v zgornji prebavni cevi.

Poročilo o primeru: V članku je predstavljen 58-letni bolnik, ki smo ga sprejeli zaradi znakov krvavitve iz zgornje prebavne cevi. V diagnostičnem postopku smo opravili nujno endoskopijo zgornje in spodnje prebavne cevi, UZ trebuha, CT-angiografijo in ileojejunografijo. Slikovne preiskave vzroka krvavitve niso pojasnile, zato smo ga odpustili. Vrnil se je po dveh tednih z znaki izrazite slabokrvnosti. Ponovljena nujna endoskopija zgornjih prebavil vzroka krvavitve ni pojasnila. Po nekaj dneh simptomatskega zdravljenja je nepričakovano umrl. Pri avtopsiji smo ugotovili sekundarno aortoduodenalno fistulo, ki je nastala po protetični rekonstrukciji trebušne aorte in bila vzrok izkrvavitve.

Zaključek: Nekateri vzroki akutne krvavitve v zgornji prebavni cevi so včasih zelo presenetljivi. Pri diagnostični obravnavi teh bolnikov in vrednotenju izsledkov se moramo zavedati omejitev slikovnih postopkov.

Abstract

Purpose: Aortoenteric fistula is an extremely rare source of upper gastrointestinal tract haemorrhage (UGIH). **Case report:** We present the case of a 58-year-old man who was admitted to our ward with UGIH. During diagnosis, urgent upper and lower GI endoscopy, abdominal US, CT angiography and ileojejunography were performed. Unfortunately, the bleeding source was not found and the patient was discharged. He returned 2 weeks later with profound anemia. Urgent upper GI endoscopy did not elucidate the bleeding source. The patient died suddenly a few days after receiving symptomatic treatment. At autopsy, a secondary aortoduodenal fistula due to prosthetic aortic reconstruction was found, which had led to fatal haemorrhage.

Conclusion: Some sources of UGIH are very rare. In the diagnostic work up of these patients we should be aware of the limitations of different imaging procedures.

INTRODUCTION

Acute upper gastrointestinal tract haemorrhage (UGIH) is a condition that is often seen in emergency medicine. The main causes are complications of peptic ulcers in the upper GI tract. Haemorrhagic erosive changes of the mucosa, ruptured varices in the oesophagus and/or stomach, and Mallory-Weiss syndrome are also frequent. Angiodysplasia, Dieulafoy's syndrome, Meckel's diverticulum and primary aortoenteric fistula are rarer (1, 2). Acute GI haemorrhage is a condition that requires in-depth clinical assessment, efficient symptomatic treatment, and urgent endoscopy to determine the causes of haemorrhage (3). Despite standard procedures of treatment for these patients and enhanced diagnostic possibilities, we sometimes face challenges that cannot be resolved in time (4).

This case report describes a patient who suffered a fatal haemorrhage due to aortoduodenal fistula caused by an aortic prosthesis, which was inserted 15 years ago because of advanced atherosclerosis at the aortofemoral bifurcation.

Case report

A 58-year-old man, who was a long-time smoker (up to 50 cigarettes a day for over 30 years), was admitted to our department due to recurrent melaena in the previous week. For the previous 20 years he had been suffering from atherosclerotic complications and a reconstructive procedure was performed on the abdominal aorta. Fifteen years ago, a Y-prosthesis at the aorto-femoral bifurcation was inserted. At clinical examination, the patient was asthenic, with anaemia, and traces of melaenic faeces were confirmed in the rectum. Haemography confirmed anaemia with an erythrocyte count of 1.94×10¹² and haemoglobin at 60 g/l. In the diagnostic procedure, urgent endoscopy of the upper and lower GI tract was performed. Upper GI endoscopy confirmed only minor traces of haematinised blood in the stomach and duodenum, erosive oesophagitis and erosive changes in the gastric mucosa. In the large bowel, traces of melaenic faeces and some diverticula were found. In the following days, abdominal US, CT angiography and ileojejunography

were performed. Imaging methods did not confirm the cause of haemorrhage and profound anaemia. During hospitalisation, the patient was treated symptomatically with infusions, iv proton pump inhibitors (pantoprazole 40 mg twice daily), iv tranexamic acid (1000 mg three times daily) and 1 l of concentrated erythrocytes. Clinically and through laboratory testing we did not register repeated haemorrhaging and the patient was discharged after 2 weeks.

The patient was readmitted 2 weeks later for anaemia with an erythrocyte count of 2.13×10^{12} and haemoglobin at 65 g/l. Urgent upper GI endoscopy did not show traces of haemorrhage or its cause. He was treated with infusions and iv proton pump inhibitors and a transfusion of 800 ml of concentrated erythrocytes. After the transfusion, he died suddenly with signs of unexplained haemorrhage. The autopsy confirmed a secondary aortoduodenal fistula that developed due to mechanical erosion of the prosthetic material into the adjacent duodenum (Figs. 1 and 2).

DISCUSSION

Aortoenteric fistula is a rare cause for acute UGIH (6-8). It represents a communication between an enteric loop and the aorta. Primary aortoenteric fistula (PAF) is a spontaneous communication between the aorta and the digestive tract, whereas secondary aortoenteric fistula (SAF) is usually a complication after reconstructive prosthetic surgery of the aorta, including aorto-bifemoral bypass implantation (9, 10). According to the literature, the occurrence of PAF in the last 10 years has not changed significantly (0.04–0.07%), but the frequency of SAF has increased in recent decades to 0.6-2.3% (10). Most primary fistulas (73%) develop from atherosclerotic aneurysms of the infrarenal part of the abdominal aorta, but those caused by an aneurysm of the thoracic aorta are rare. In approximately 26% of cases, they form after injury or because of a mycotic aneurysm. Other rare causes include radiation injury and malignant or infectious diseases in the abdominal cavity (10). Most SAFs originate in the infrarenal part of the abdominal aorta, and the most frequent causes are aneurysm or surgical aneurysm repair with prosthetic implants. Aortoenteric fistulas most often develop in the duodenum, which crosses the aorta (horizontal part, 57%); other areas of the duodenum are less frequently affected (9%) (12). These fistulas form also in other parts of the digestive tract, in the small (8%) and large (4%) intestine and the stomach (4%). Most surgeons share the opinion that SAF formation is due to mechanical erosion of the wall of the duodenum with aneurysmatic blood vessels or implants (5, 9). Others believe that perioperative infection of the prosthesis leads to the formation

of pseudoaneurysms, which by pressure, lead to a connection with the GI lumen. One of the possible explanations is the formation of a periprosthetic fistula that arises at the point of the haematoma by the suture line. In later course, infection develops most probably from the intestine, which enables the formation of the connection between the aorta and the digestive tract.

Clinically, in these patients, haemorrhage is the most common sign followed by abdominal pain, which

can radiate to the lower back and kidney area (9). A pulsating mass in the abdomen is one of the rarest signs, which occurs in about 20% of patients. A frequent symptom is sudden and unexplained fever caused by the migration of microorganisms from the intestine directly into blood vessels. Among the diagnostic procedures, urgent endoscopy of the upper digestive tract is the most appropriate method for determining the cause of haemorrhage, although it has its limitations. The endoscopist can only indirectly conclude the cause of haemorrhage, if the blood is flowing from the horizontal part of the duodenum. The sensitivity of these diagnostic methods is supposed to be 25-80% (10, 11). CT angiography is among the most accurate methods for confirming aortoenteric fistula, with a sensitivity up to 93%, and at the same time, has an advantage over endoscopy because it is fast, non-invasive and highly predictive. Angiography of abdominal blood vessels has its limita-



Fig 1. Aortoduodenal fistula.



Fig 2. Aortoduodenal fistula.

tions. As a result of the blood clot that forms in the fistula,

angiography often does not confirm the communication between the digestive organ and the aorta. Abdominal US has its limitations in this life–threatening condition and is not recommended.

The treatment of patients with aortoenteric fistula is usually operative and depends on the aetiology of the fistula. In the literature, different techniques of surgical repair have been described: type A, blind closure of the aorta, removal of the prosthesis, and construction of an extra-anatomical bypass; type B, partial substitution of the affected part of the prosthesis covered by omental sleeve; and type C, total removal of the prosthesis without reconstruction (12). Surgery is associated with high mortality of 20–93% (9, 11). Recently, there has been accumulating data about insertion of intraluminal prostheses by interventional radiologists (8, 10). Without operative correction, the fistula can lead to septic complications (9, 12).

In the current case, we could not confirm aortoenteric fistula, despite the repeated diagnostic procedures and consideration of this aetiology. This case higlights the importance of clinical awareness in patients with unexplained GI haemorrhage after reconstructive prosthetic aortic surgery.

CONCLUSION

The treatment of patients with acute UGIH is usually performed in accordance with standard procedures. In confirming the aetiology, we must allow the possibility that causes are uncommon and that the results of diagnostic procedures can be misleading. In patients with signs of GI haemorrhage and known aneurysm or reconstructive surgery of the abdominal aorta, we also have to consider the formation of aortoenteric fistulas, which should be confirmed or disproved using an appropriate diagnostic procedure. At this point, we have to be aware of the limitations of diagnostic imaging methods.

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