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CASE REPORT

# Post-bulbar duodenal ulcer with anterior perforation with kissing ulcer and duodenocaval fistula: A case report and review of literature

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#### Abstract

# **BACKGROUND**

A post-bulbar duodenal ulcer (PBDU) is an ulcer in the duodenum that is distal to the duodenal bulb. PBDU may coexist with a synchronous posterior ulcer in rare occurrences, resulting in a kissing ulcer (KU). Duodenocaval fistula (DCF) is another uncommon but potentially fatal complication related to PBDU. There is limited knowledge of the scenarios in which PBDU is complicated by KU and DCF simultaneously.

#### CASE SUMMARY

A 22-year-old man was admitted to the emergency department with abdominal pain, stiffness, and vomiting. The X-ray showed pneumoperitoneum, suggesting a perforated viscus. Laparotomy revealed a KU with anterior perforation and a DCF. After Kocherization, venorrahphy was used to control caval bleeding. Due to the critical condition of the patient, only primary duodenorrahphy with gastrojejunostomy was performed as a damage control strategy. However, later, the patient developed obstructive jaundice and leakage, and two additional jejunal perforations were detected. Due to the poor condition of the duodenum and the involvement of the ampulla in the posterior ulcer, neither primary repair nor pancreatic-free duodenectomy and ampull-oplasty/ampullary reimplantation were considered viable; therefore, an emergency pancreaticoduodenectomy was performed, along with resection and anastomosis of the two jejunal perforations. The patient had a smooth recovery after surgery and was discharged after 27 d.

# **CONCLUSION**

The timely diagnosis of PBDU and radical surgery can aid in the smooth recovery of patients, even in the most complex cases.

**Key Words:** Duodenal ulcer; Duodenocaval fistula; Kissing ulcer; Emergency Whipple's



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surgery; Case report

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Core Tip: A post-bulbar duodenal ulcer, in combination with a kissing ulcer (KU) and a duodenocaval fistula (DCF), is a severe complication with a high mortality rate. In the present case, the patient had a KU with anterior perforation and a DCF. After Kocherization, venorrahphy was used to control caval bleeding. The patient, however, later developed obstructive jaundice and leakage. Due to the poor condition of the duodenum and the involvement of the ampulla in the posterior ulcer an emergency pancreaticoduodenectomy was performed, along with resection and anastomosis of the two jejunal perforations. The patient had a smooth recovery after surgery.

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#### INTRODUCTION

According to current estimates, post-bulbar duodenal ulcers (PBDU) account for 5%-10% of all duodenal ulcers[1]. Most of these ulcers develop in the second part of the duodenum, with the majority occurring on the posteromedial wall of the duodenum[2]. The overall incidence of duodenal ulcers is 9.33%[3]. According to autopsy studies, 5%-20% of peptic ulcers are post-bulbar ulcers, whereas 5% of duodenal ulcers are situated distal to the bulb[4]. The clinical presentation and specific diagnostic criteria for PBDU have not yet been fully established, making diagnosis and treatment difficult[5]. Abdominal pain is the most common peptic ulcer symptom; however, melaena and hematemesis are also common[6].

PBDU, in a rare condition, can accompany a synchronous posterior ulcer and form a 'kissing' ulcer (KU)[7-10]. In the context of kissing duodenal ulcer, some may choose a vagotomy and pyloroplasty over a proximal gastric vagotomy. However, plication therapy for perforated duodenal ulcers results in postoperative bleeding problems due to KU. Bleeding in the upper gastrointestinal tract makes duodenal bulb ulcers worse and less responsive to treatment[11]. Any instance of symptomless hematemesis should be thoroughly explored for post-bulbar ulcers. Furthermore, since these abnormalities may overlap with presentations commonly seen in Zollinger Ellison syndrome (ZES), a precise diagnosis and treatment strategy become critical[12].

Another severe and rare complication of PBDU is the duodenocaval fistula (DCF). DCF is an uncommon but severe form of digestive fistula. It develops due to problems with the duodenuminferior vena cava junction. Nontraumatic DCF is sporadic and may be caused by various factors such as penetrating duodenal peptic ulcers, foreign bodies, malignancies, right nephrectomy, and radiation therapy to the upper abdomen. DCF is often distinguished by gastrointestinal bleeding; however, it can also be accompanied by fever and infection. DCF should be approached cautiously, and decisions should be made quickly since it is associated with a high mortality rate before surgical intervention[13].

Several cases on the treatment of PBDU and when KU or DCF complicates PBDU have been documented in the literature. However, PBDU in conjunction with KU and DCF is not adequately described in the literature. This study presents the successful management of a rare case of PBDU complicated by KU and DCF, providing a detailed overview of diagnostic problems and surgical complications. A brief account of the previous research published in this field is also included.

#### **CASE PRESENTATION**

# Chief complaints

Stomach discomfort and vomiting for two days.

#### History of present illness

On January 24, 2019, a 22-year-old man came to the emergency department with stomach discomfort and vomiting for two days. He appeared to be stressed due to acute dehydration.

#### History of past illness

The patient was taking non-steroidal anti-inflammatory drugs with no history of peptic ulcer disease. He was hypotensive and tachycardic, with blood pressure (BP) of 100/70 mmHg and a pulse rate of 110 beats per minute.

# Personal and family history

There was no family history relevant to this case.

### Physical examination

Physical examination indicated board-like stiffness of the abdomen, and auscultation revealed a negative bowel sound.

#### Laboratory examinations

See Table 1 for lab results at admission.

#### Imaging examinations

An erect chest radiograph revealed air under the diaphragm.

#### FINAL DIAGNOSIS

Following resuscitation, a nasogastric tube (NG) and a foley catheter were placed, and the patient became anuric. He underwent an exploratory laparotomy, which revealed a KU, a post bulbar ulcer in the second part of the duodenum with a severely deformed and fibrotic duodenum, and an anterior ulcer that was perforated and obstructed by clots (Figure 1, Video 1).

#### TREATMENT

Due to the penetration of the posterior ulcer into the inferior vena cava (DCF), which was in contact with the ampulla, the duodenum was filled with venous blood. Duodenotomy was performed and pressure was applied to achieve temporary control of the source until the blood transfusion. Duodenal Kocherization was performed and after proximal and distal control of the inferior vena cava with sponge-on-sticks, caval hole venorrahphy was conducted. The patient received the equivalent of 2 total blood volumes of packed red blood cell, platelets, and fresh-frozen plasma. Due to the critical condition of the patient, a damage control approach was used. Gastrojejunostomy was conducted after primary repair (transverse duodenorrahphy/duodenoplasty of the duodenotomy that included both ulcers) as a bypass procedure due to narrowing of the duodenal. A drain was left in the subhepatic space, and the patient developed obstructive jaundice and biliary leakage from the drain while in the intensive care unit (ICU). He was brought for re-exploration, which revealed two additional perforations in the jejunum. Due to the poor condition of the duodenum and the involvement of the ampulla in the posterior ulcer, neither primary repair nor pancreas sparing duodenectomy and ampulloplasty/ampullary reimplantation were viable options. An emergency Whipple procedure (pancreaticoduodenectomy) was performed, along with resection and anastomosis of the jejunal perforation.

# **OUTCOME AND FOLLOW-UP**

The patient recovered quickly from the surgery. However, he experienced two episodes of aspiration pneumonia, for which he was intubated and ventilated in the ICU. The dehiscence of the wound developed as a result of renal failure. On the eighth postoperative day, a computed tomography (CT) scan revealed no collection or leakage. Every other day, the drain effluent was sent for analysis of amylase and bilirubin, and white cell count and only turbid fluid was found, without amylase leakage or pus. On the  $12^{th}$  postoperative day, the patient was extubated and kept in the ICU with 2 Liters of  $O_2$ through a nasal cannula saturating 98%, with vitals of BP: 137/70 mmHg, pulse: 104 beats per minute, and temperature: 37.8 °C. The last culture from the chest revealed Klebsiella pneumonia, and Vancomycin-resistant enterococci. Other laboratory findings are given in Table 1.

On day 13 after the operation, the patient was fed through NG and tolerated well glasgow coma scale of 13/15. Evaluation of anastomotic leakage (enteric, biliary, or pancreatic) by abdominal CT revealed no leak or collection. Tigecycline, Colistin, and Imipenem were prescribed to the patient. After 27 d, the patient was discharged with a good clinical condition.

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Table 1 Laboratory parameters of the patient at admission	
Parameter	Value
Hite blood count	19
Hemoglobin	9.2
Creatinine	230
Urea	15
Sodium	146
Albumin	31
Alanine transaminase	29
Aspartate transferase	42
Total bilirubin	30



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Figure 1 Exploratory laparotomy revealed post-bulbar duodenal ulcer. It can be seen that the anterior ulcer perforation is plugged with a clot.

#### **DISCUSSION**

Early diagnosis and successful management of PBDU present several challenges. The risk of mortality and morbidity increases many folds when KU and DCF complicate the condition; however, little is understood about the clinical signs, diagnosis, and management when all these complexities (PBDU, KU, and DCF) arise simultaneously. This study unveiled numerous critical aspects of this complex condition by offering a comprehensive discussion on the successful treatment of a patient with PBDU, KU, and DCF. This case presented several diagnostic and clinical challenges, and the patient was subjected to surgical treatment with caval venorrahphy, pancreaticoduodenectomy, segmental enterectomy, and jejunojejunostomy, which resolved KU and DCF and jejunal perforations.

Perforations in the duodenum can be free or limited. The term "free perforation" refers to the time when the intestinal material seeps into the abdominal cavity, resulting in diffuse peritonitis. Limited perforation occurs when an ulcer produces a full-thickness hole, but the open leaking is blocked by surrounding organs such as the pancreas. Although patients with duodenal perforation require surgical treatment in most cases, in patients with perivaterian injuries, conservative management can also produce successful outcomes. In acute duodenal perforations, Whipple surgery is highly complicated [14,15]; therefore, in our case, radical surgery differed initially. However, when pancreatic-free duodenectomy and ampulloplasty/ampullary reimplantation became unfeasible, emergent Whipple surgery was successfully performed, reflecting the feasibility of radical approaches in the treatment of such complex cases.

The most common indication for surgery is bleeding, and surgical options include stomach resection +/- vagotomy. The mortality rate is 2.6%, except in fistulous cases, which have a mortality rate of 7.7%. Of note, when complicated with KS, the mortality rate in bleeders is reported to be as high as 50% [16]. Therefore, in events of gastrointestinal blood loss in a perforated duodenal ulcer, an intraoperative search for a posterior KS should be considered. If a KU was found, an acid-reducing operation and suture ligation are viable approaches[7].

Compared to bulbar and gastric ulcers, the frequency of bleeding in PBDU is approximately twice as high. PBDU appears later in life and affects men more than women[17]; however, unlike bulbar duodenal ulcers, patients with PBDU tend to show severe acidity, a persistent pattern of acid secretion, and different clinical and radiographic findings[18]. Postoperative mortality from PBDU is greater than bulbar duodenal ulcer[3]. In a study from India that provided a detailed description of the aspects associated with duodenal ulcer, post-bulbar ulcer vs bulbar ulcer was found to be 1:1.5, with deformed bulbs observed in half of the cases[17]. Smokers accounted for 42% of patients with duodenal ulcers, while tobacco chewers accounted for 15% and alcoholism for 18%. Consumption of tea, rice, and spices has also been associated with increased acid secretion and the development of duodenal ulcers. Most importantly, H. Pylori was estimated to infect 80% of the population. It may be noted that, unlike duodenal bulb ulcer, in certain cases, PBDU did not heal with Helicobacter pylori eradication therapy, suggesting that post-bulbar ulcer etiologically differs from bulbar ulcer[19]. Notably, H.pylori was also implicated in the development of KU in the duodenal bulb[9].

In 1989, the first recorded instance of a duodenal ulcer associated with pentagastrin-fast achlorhydria was described[20]; wherein a 55-year-old male was diagnosed with a post bulbar duodenal ulcer, hemorrhage, and fasting hypergastrinemia, and ulcer healing was reported in eight weeks, after antrectomy, vagotomy, gastrojejunostomy, and a course of sucralfate medication. In another study, 12 patients were reported to have PBDU[21]. In ten patients, a truncal vagotomy and outlet surgery were performed and the anterior duodenotomy extended to the stenotic region. A Jaboulay gastroduodenostomy was performed in one patient, while a pyloroplasty duodenoplasty was performed in another. There were no deaths among these individuals during the six-year follow-up period. However, three individuals had recurrences of peptic ulcers, which could be related to undetected post-bulbar stenosis after surgery. This showed that to detect the ulcer, the duodenum must be checked by intraluminal palpation in all patients undergoing surgery for peptic ulcer disease.

Notably, in 121 individuals with PBDU, 72.7% had duodenal stenosis, 41.3% had penetration, and 5.8% had choledochoduodenal fistulas [22]. In approximately 40% of the patients, the progress of the ulcer was exacerbated by bleeding and in 8.3% by perforation. In 34 patients, a selective proximal vagotomy was performed, and in 28 cases, a selective proximal vagotomy and a draining operation were performed. In 41 patients, proximal vagotomy and duodenoplasty were performed. The use of selective proximal vagotomy and duodenoplasty produced better results. There were no deaths or serious complications. In contrast, a comprehensive case review of 1087 patients with PBDU showed complications in 1014 of them; importantly, penetration occurred in 707 individuals in conjunction with hemorrhage from stenosis[23]. Organ saving techniques combined with vagotomy have been shown to offer benefits over resection techniques[24]. After surgery, therapeutic outcomes can vary depending on the patient's predisposition to the post-bulbar lesion[16].

Our patient also had DCF, a type of intestinal fistula that is a fatal clinical entity with a high death rate prior to definitive therapy [13]. If DCF is clinically suspected, the first-line study should be CT and magnetic resonance imaging, with a thorough evaluation of images of inferior vena cava (IVC) and surrounding structures. The prognosis is decided by early diagnosis and surgery before bleeding starts. DCF treatment includes fistula closure, as well as duodenal and IVC repair[25]. This argument is based on a postmortem examination of a 54-year-old man, which found that the cause of death was upper gastrointestinal hemorrhage, resulting in a considerable volume of blood in the intestinal lumen due to gastric ulcer rupture in the IVC. In our case, DCF management involved surgical intervention, which was decided after a meticulous assessment of the extent of damage to the duodenum and IVC. Barloon et al[26] reported a morbidity rate of 61% after radical surgery. To prevent fistula recurrences, surgerybuttressed repair, such as a jejunal patch or an epiploic flap, is generally preferred; however, other approaches, such as truncal vagotomy, antrectomy, and/or duodenal exclusion, can be utilized in combination, particularly in the case of a peptic ulcer. Conservative surgical therapy was also successfully applied to a 73-year-old man with DCF, occult intestinal bleeding, and sepsis [27]. In this case, the IVC and duodenum were sutured after sharp dissection, duodenal mobilization, and digital control of hemorrhage. The duodenal exclusion was performed using gastroenterostomy and truncal vagotomy of antral stapling and interposition of the epiploic patch to prevent recurrence of the fistula [27].

The presence of a gastric ulcer with a fistula in the IVC and food embolization in the lung has also been documented, indicating the severity of the condition[28]. The first case of DCF caused by a large descending duodenal peptic ulcer was described in 1990[29]. DCF hemorrhage was contained by direct compression above and below the fistula in another example involving a 49-year-old male, and the IVC defect was repaired with 5/0 prolene[30]. In 1996, a case of polymicrobial fungemia and fatal gastro-Intestinal hemorrhage associated with DCF caused by a peptic ulcer was reported. This raised the possibility of candidal endocarditis[31]. The first case of DCF with peptic ulcer showed complications due to embolization of the intestinal contents in the lung, with numerous intravascular mucin[32]. In 2005, a case of a 44-year-old patient with DCF was also reported. The patient had no history of peptic ulcer disease. Septic shock preceded hemorrhagic shock as a clinical characteristic, but only after laparotomy was the diagnosis established [33]. Finally, in our case, the patient developed jaundice, which is rare, with a prevalence of 0.14% [34]. The development of jaundice is due to the close relationship of the common bile duct with the second part of the duodenum. It may be noted that though our case provides vital information on the management of PBDU, KU, and DCF, more studies are needed to standardize the diagnostic and therapeutic approaches.

# CONCLUSION

PBDUs generally involve a surgical emergency and are difficult to diagnose and manage. This rare case highlights that PBDU should be the first of surgical intent and a radical procedure should be considered if viable. In our case, only primary repair of the two ulcers was performed along with a proximal drainage procedure (gastrojejunostomy); however, when additional perforations were detected and conservative measurements were not viable, emergent Whipple surgery was successfully performed. The duodenocaval fistula, in particular, is difficult to diagnose in this situation and is linked with a significant mortality rate before conclusive therapeutic efforts. Our case demonstrates that a careful diagnosis and timely treatment will be helpful for patient recovery and a good prognosis.

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# **FOOTNOTES**

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