Case Report

Massive upper gastrointestinal hemorrhage due to adenosquamous carcinoma of the pancreas: Case report and literature review

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Background

Adenosquamous carcinoma (ASC) is an uncommon primary tumor of the pancreas comprising only 1% of all pancreatic malignancies. Due to its rare nature, diagnosis is frequently made at the time of surgery or autopsy. Widely disseminated disease is often present at the time of diagnosis, making surgical resection challenging, if not contraindicated, and conferring a generally poor prognosis. We present the case of a 57 year-old male with adenosquamous carcinoma of the pancreas, complicated by massive upper gastrointestinal hemorrhage, and offer a brief discussion of the pertinent literature.

Case report

A 57 year-old Hispanic male with diabetes, hypertension and chronic kidney disease presented to the emergency department with complaints of dizziness, fatigue, night sweats, unintentional weight loss and melena for the past three months. He was found to be profoundly anemic (hemoglobin 7.3 g/dL) with a slight leukocytosis (12,800/μL). He was admitted to the hospital, resuscitated with several liters of crystalloid solution, transfused two units of packed red blood cells and placed on a continuous intravenous proton pump inhibitor infusion. Upper endoscopy was performed on hospital day three and five, both of which revealed the presence of a large, multi-lobulated tumor within the gastric lumen (Fig 1). The mass showed several areas of deep ulceration and continuous hemorrhage. The first endoscopy revealed a friable mass within the stomach, which contained multiple areas of active hemorrhage. Several biopsies were taken and epinephrine was injected in an attempt to control bleeding. The patient was given two additional units of packed red blood cell transfusions and intravenous crystalloid solution. Upon repeat endoscopy 72 hours later, uncontrollable, spontaneous bleeding was noted from several ulcerated areas and the patient became hemodynamically unstable. At this point, the procedure was aborted, the patient was rapidly intubated and taken to the ICU for stabilization. The estimated blood loss for both endoscopies was approximately one liter of blood.

On hospital day six the patient underwent a CT scan which confirmed the presence of a 10 cm × 10 cm multi-lobulated mass within the lumen of the stomach which was abutting the tail of the pancreas (Fig 2). Foci of central necrosis were observed on one side of the lesion but there was no evidence of obvious metastatic disease. The next morning, he was taken to the operating room for exploration. Intraoperatively, a large mass involving the posterior wall of the stomach and the lesser sac was identified. There were multiple areas of profuse hemorrhage along the surface of the tumor. Surgery was initially attempted laparoscopically, but had to be converted to a laparotomy in order to safely separate the mass from the anterior

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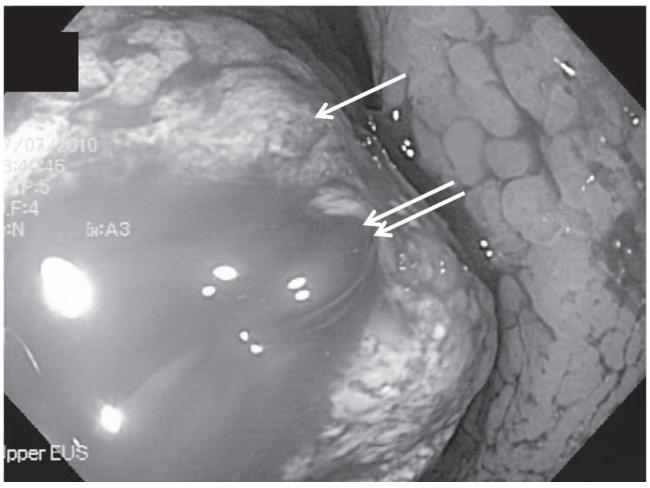

Figure 1. Upper endoscopy revealed the presence of a large, ulcerated, multi-lobulated mass (arrow) within the gastric lumen. The mass showed evidence of profuse hemorrhage (two arrows), which was temporized by epinephrine injection.

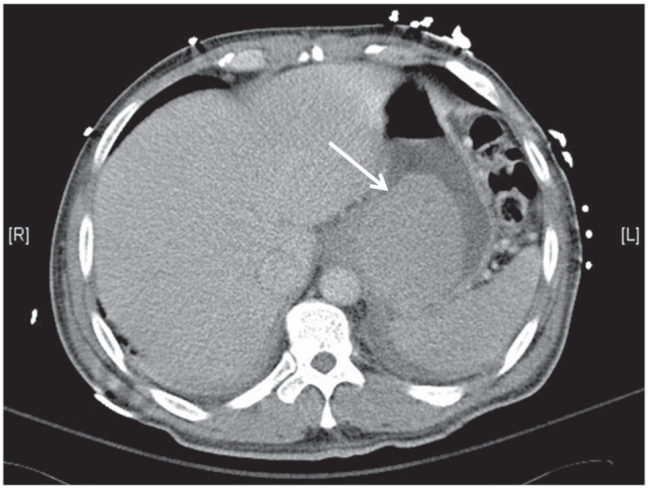

Figure 2. CT scan showed a 10 cm x 10 cm multi-lobulated mass (arrow) located within the gastric lumen which was inseparable from the tail of the pancreas. Several foci of central necrosis were observed within the mass.

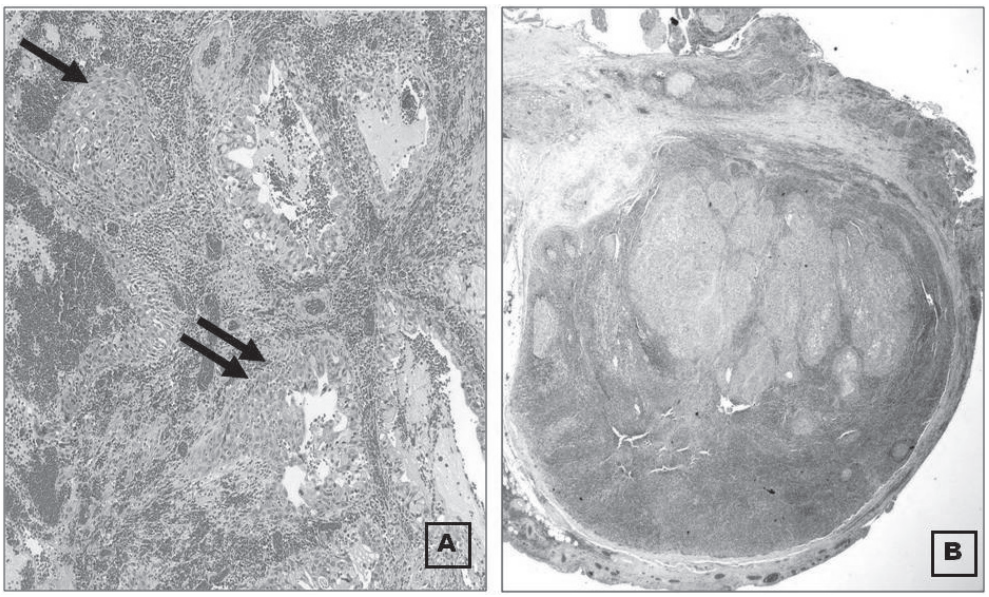

Figure 3 (A). Adenosquamous carcinoma composed of malignant squamous elements (arrow) and glandular elements (two arrows); (B). Lymph node with metastatic malignant squamous carcinoma.

surface of the pancreas and adjacent retroperitoneum. This separation was performed bluntly in order to gain control of the ongoing hemorrhage. Since the lesion extended superiorly towards the gastroesophageal junction, a thoracoabdominal incision was made to obtain a sufficient proximal margin. Ultimately, the patient underwent a total gastrectomy, roux-en-Y esophagojejunostomy and feeding jejunostomy tube placement. A total of 2.5 liters of blood was lost intraoperatively and the patient was given six units of packed red blood cells and four units of fresh frozen plasma during surgery. Frozen section diagnosis was consistent with invasive adenosquamous cell carcinoma. Upon final pathological examination, the tumor was conclusively determined to be of pancreatic origin. Extensive sampling and permanent sections of the tumor revealed predominantly high grade adenosquamous cell carcinoma with marked nuclear atypia and necrosis and focal malignant glandular elements, consistent with adenosquamous carcinoma (Fig 3). Two of 13 perigastric lymph nodes were positive for metastatic disease, composed of only the squamous component.

The patient had an uneventful post-operative course and was discharged home on post-operative day 17. He remains well one month after surgery and has been referred

to radiation and medical oncology. After he sufficiently recovers from surgery, consideration will be made to pursue potentially curative resection versus chemoradiation or palliative treatment.

Discussion

Adenosquamous carcinoma of the pancreas, also referred to as "adenocanthoma" and "mucoepidermoid carcinoma" (1,2) is a rare entity, representing only 1-4% of all known pancreatic malignancies (3-5). Similar to adenocarcinoma, the most common symptoms associated with ASC are weight loss, anorexia, malaise, abdominal pain, fatigue and nausea. These tumors possess components of both glandular and malignant squamous cell origins, which should both be present to ensure proper diagnosis. Diagnosis of ASC is challenging and frequently not made until the time of surgery or during post-mortem examination. The use of ultrasound-guided FNA or ERCP-guided aspiration for making a pre-operative diagnosis remains controversial. No imaging criteria are specific to ASC, causing most of these tumors to be mistaken for ductal adenocarcinoma of the pancreas when initially imaged. However, CT findings of large pancreatic lesions with infiltration of surrounding tissues and central necrosis have been reported and should raise one's suspicion for ASC (6).

Widely disseminated disease is commonly present at the time of initial presentation. Although diffuse disease usually prevents resection, palliative surgery may still be indicated in cases of hemorrhage, perforation or obstruction.

Even when potentially curative surgical resection is performed, prognosis is generally regarded as extremely poor. In 2008, Okabayashi et al reviewed 39 cases of ASC treated with pancreatic resection (pancreaticodudenectomy, distal pancreatectomy or total pancreatectomy) spanning 1980 through 2007 (7). In this review, the 1-year and 3-year survival rates following pancreatic resection were 25% and 14%. Nevertheless, some reports have demonstrated a survival benefit for patients undergoing an R0 resection (8). Similarly, Voong et al showed a survival benefit for patients treated with adjuvant chemoradiation therapy (4).

While direct invasion of adjacent organs has been reported for ASC (5), it usually does not lead to erosion through the wall of hollow viscera. When present, this presents an obvious challenge to the surgeon, but should not be interpreted as a contraindication to surgical resection. Gastrointestinal bleeding has been associated with tumors of multiple abdominal organs, but rarely so with primary lesions of the pancreas. Several treatment strategies for malignancy-related GI bleeding are commonly employed, including endoscopic techniques, angiographic embolization and surgical therapy (9). All modalities are useful in the appropriate setting, but treatment must be individualized for each case.

In conclusion, we present a case of a 57 year-old male with adenosquamous carcinoma of the pancreas. Our case is unique in that we consider it to be the first that presented as a massive, acute upper gastrointestinal bleed after erosion through the posterior gastric wall. This case illustrates an atypical presentation for this disease, forcing us to heighten our awareness of these lesions in order to ensure prompt diagnosis in future cases.

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