Gastrovesical fistula as a rare complication following endoscopic transluminal drainage of walled-off necrosis—a case report

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Background: This study highlights an unusual and previously unreported adverse event (AE) following the minimally invasive treatment of pancreatic walled-off necrosis (WON). The standard treatment for WON currently involves primary drainage via an ultrasound-guided endoscopic, typically transgastric, approach. This method is associated with lower mortality and morbidity rates compared to traditional surgery. However, emerging AEs from these procedures may necessitate the involvement of a multidisciplinary team. Our case highlights the potential for gastrovesical fistula development as a rare AE following endoscopic drainage. Treatment for our patient prioritized individualized and non-surgical strategy, although surgical revision was also considered.

Case Description: A 42-year-old male presented with a large symptomatic pancreatic WON refractory to conservative management, necessitating transgastric drainage. Despite the gradual evacuation of the WON contents, treatment was complicated by stent-related issues, including inadvertent bladder penetration. Rather than surgical correction, a collaborative approach among urology, gastroenterology, and surgery teams was employed, focusing on conservative treatment strategies. This approach successfully resolved the fistula, leading to the patient's full recovery.

Conclusions: Given the increasing use of endoscopic transluminal drainage in (peri)pancreatic collections, it is crucial to be aware of all potential AEs. To our knowledge, this is the first documented case of gastrovesical fistula following drainage of WON. Early recognition and a multidisciplinary approach are vital to manage this event.

Keywords: Necrotizing pancreatitis (NP); walled-off necrosis (WON); endoscopic drainage; gastrovesical fistula; case report

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Introduction

Acute pancreatitis (AP) constitutes a medical challenge, often necessitating a multidisciplinary approach and a spectrum of measures spanning from conservative therapy to surgical interventions (1,2). The Revised Atlanta Classification distinguishes mild [no organ failure (OF) and no local complications], moderate (transient OF resolving in <48 hours or local complications), and severe forms (persistent OF lasting ≥48 hours with or without local complications) (3). While mild AP is typically selflimiting and permits patients to leave the hospital within a few days, severe AP is associated with protracted hospitalization and high mortality. The pathophysiological correlate of local complications is primarily necrosis of the (peri)pancreatic tissue [necrotizing pancreatitis (NP)] which occurs in approximately 15-20% of patients (1,3-6). Walled-off necrosis (WON) evolves as a sequel to acute necrotic collections. These encapsulated necrotic areas can lead to further clinical deterioration, with mortality rates reaching up to 39% in cases of infected necrosis (1,3-6). As such, drainage of necrotic contents is a crucial step when indicated.

Surgical necrosectomy was once considered the gold standard. However, minimally invasive methods, including endoscopic ultrasound (EUS)-guided transluminal drainage (ETD) and percutaneous approaches, have gained

Highlight box

Key findings

 Our case-report describes a multidisciplinary approach to the treatment of a very rare (to our knowledge, the first documented case) complication of endoscopic drainage of walled-off necrosis, which is gastrovesical fistula.

What is known and what is new?

- So far this complication—at least to our knowledge—has not been described in the world literature.
- The manuscript describes a comprehensive multidisciplinary
 approach to the management of a rare complication of acute
 pancreatitis, gastrovesical fistula. Its therapy is now being
 encountered by specialties that have not previously come into
 contact with the disease at all, and are forced to take an active role
 in its treatment.

What is the implication, and what should change now?

 New modern methods bring new, previously unknown complications, for which we must—in all specialties—be prepared to address them proactively. prominence due to their reduced invasiveness and potential to limit adverse events (AEs) (2,5). Surgical interventions are now primarily reserved for cases where other measures prove ineffective and/or unfeasible (7). This paradigm shift reflects the evolving trend towards optimizing patient outcomes while minimizing the associated risks of surgery. Nonetheless, the endoscopically centered approach is not without its risks. We present a unique case of gastrovesical fistula resulting from ETD of WON. To our knowledge, this is the first documented case of such an event. We present this case in accordance with the CARE reporting checklist (available at https://acr.amegroups.com/article/view/10.21037/acr-24-18/rc) (8).

Case presentation

A 42-year-old male, height 182 cm, weight 75 kg, body mass index (BMI) 22.4 kg/m², was admitted to the internal medicine department for diarrhea, weight loss, subfebrile (37.2 °C). The discomfort lasted approximately 2 weeks. He had a personal history of chronic pancreatitis and hepatopathy of toxonutritive etiology and an uncomplicated gastric ulcer that had resolved, with no history of abdominal surgery. He had no known allergies, was an active smoker, and a former alcoholic. He was chronically medicated with proton pump inhibitors (PPIs) and analgesics only. He worked as a carpenter. Initial laboratory examination showed leukocytosis (19×10⁹/L) and elevation of C-reactive protein (CRP) (220 mg/L). Liver function tests were normal, serum alpha-amylase (AMS) was elevated (225 U/L), as was serum lipase (LPS) (183 U/L). Computed tomography (CT) scan with contrast agent revealed an extensive pancreatic WON (Figure 1). On physical examination, the patient was subfebrile and had only mild palpatory abdominal pain in the epigastrium. He was indicated for ETD of the necrotic collection. During the procedure, a 20 mm × 16 mm lumenapposing metal stent (LAMS) (Hanarostent, M.I. Tech, Seoul, South Korea) was inserted through the stomach wall into the WON, and a coaxial 10 Fr × 5 cm doublepigtail plastic stent (DPS) (Advanix; Boston Scientific, Marlborough, MA, USA) was then placed through the LAMS (Figure 2). The patient was secured with antibiotics, the subsequent course of hospitalization was uncomplicated, follow-up CT scan showed significant regression of the collection (Figure 3). The patient was discharged to home care after calming down and normalization of laboratory results.

One month after discharge, he was readmitted to

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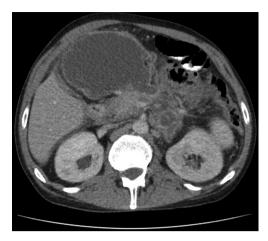


Figure 1 Initial computed tomography scan demonstrating peripancreatic necrotic collections prior drainage.



Figure 2 Lumen-apposing metal stent with a coaxially placed double pig-tail plastic stent on fluoroscopy during endoscopic retrograde cholangiopancreatography.

the Internal Medicine Clinic for febrile illness. Further endoscopic examination showed obturation of the original LAMS stent with tissue detritus, the stent was patent and the original DPS was replaced with a longer, $10~{\rm Fr} \times 7~{\rm cm}$ (Advanix; Boston Scientific). Because of continued febrile disease, a second $10~{\rm Fr} \times 5~{\rm cm}$ DPS (Advanix; Boston Scientific) was inserted into the necrotic cavity at follow-up endoscopy along with the removal of the LAMS. Follow-up CT showed regression of collections around the pancreas. The patient was discharged in good condition 3 weeks later with DPSs left *in situ*. A follow-up CT scan

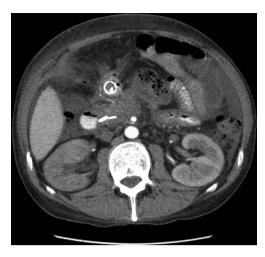


Figure 3 Follow-up computed tomography scan after successful drainage of the WON collection. WON, walled-off necrosis.

in another 3 months showed a new fluid collection in the small pelvis presacrally and dilatation of the calyceal system of the right kidney. The clinical condition was favorable, and a control endoscopy was performed with adjustment of the stent position. Urological examination with renal scintigraphy was added for dilatation of the right ureter, which showed incomplete obstruction with partial involvement of the kidney of unclear etiology. Cystoscopy with a plan to drain the right kidney was unsuccessful due to swelling and unclear mucosal changes at the mouth of the right ureter. Cystourethrography (CUG) was without pathological findings. Control cystoscopy with bladder mucosa biopsy excluded malignancy. Due to the absence of clinical complaints, the patient did not pursue the urological findings further.

In 4 months without clinical problems of the patient, the communication of the collections in the retroperitoneum and pelvis with the bladder was described for the first time during the next CT scan (Figure 4). Thus, another cystoscopy was added, during which a double J (DJ) stent (OptiSoft, Optimed, Ettlingen, Germany) was successfully inserted into the right kidney with evacuation of purulent urine; no clear fistula or stent was seen in the bladder wall, only indirect signs of fistula (erythema, irregular bladder mucosa) were indicative of the latter. Control transgastric endoscopy with stent injection showed no fistula into the bladder, only a small collection around the bladder, and for obturation of the stent draining this collection (Advanix 10 Fr × 7 cm), it was replaced with a new one. A follow-up CT scan was performed 2 weeks later, which described

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Figure 4 Follow-up computed tomography scan showing a fistula and a plastic stent within the urinary bladder.

progression of fluid collections around the bladder with its compression, gas bubbles around the bladder on the right, and probable inflammatory involvement of the bladder wall. Further cystoscopy was not indicated by the urologist at this stage due to the risk of iatrogenic bladder perforation and the development of a chronic intractable retroperitoneovesical fistula. Subjectively, the patient's condition was uneventful, CRP was low (40 mg/L) even without antibiotic therapy, therefore the patient was only clinically followed.

For the next 8 months, the patient was free of problems, regularly monitored clinically and laboratory, control CT scan was stationary, and control endoscopy by transgastric means was also favorable. Follow-up urological examination and additional CUG at this stage was favorable; CUG findings were within normal limits. A planned follow-up CT scan of the abdomen revealed a new clear presence of the longer DPS in the bladder wall, fluid collections around the bladder were stationary. The patient developed abdominal pain at this stage, was repeatedly examined in the surgical and urological outpatient clinics, inflammatory markers were low, the surgeon ruled out abdominal sudden stroke, and the urinary findings were treated with targeted antibiotics according to culture. During the control transgastric endoscopy, both DPSs were replaced with shorter ones (Advanix 7 Fr × 5 cm). Furthermore, the patient's clinical complaints were minimal. Surgical or urological revision surgery with any radical solution was repeatedly contraindicated because of the high risk of further intractable complications. A follow-up surgical examination, including CT scan, was added at 1-month intervals for febrilia, which described stationary collections around the bladder, without clear penetration of the pigtails

into the bladder, and a new collection in the abdominal wall. The latter was indicated for surgery, however the procedure entailed only incision and drainage under local anesthesia. The urologist performed a control cystoscopy, a DJ stent was replaced in the right kidney and the bladder was drained with a permanent urinary catheter. This was retained after the patient was discharged.

The patient's further course is now uncomplicated, and urological examinations at 2 and 6 months after the last hospitalization, including CUG have ruled out further complications or persistent fistula. Due to the involvement of the right kidney, which could be a source of septic complications in the future, a right-sided nephrectomy was proposed, but the patient is still undecided. Based on negative CUG, a permanent catheter was extracted. As of now the patient is subjectively completely free of discomfort, without elevated temperature or laboratory signs of inflammation. Follow-up endoscopic examination 6 months after discharge was normal, both transgastric DPSs have been replaced with new ones, and another follow-up endoscopic examination is planned in 6 months, when stents will be extracted in case of a favorable CT scan.

All procedures performed in this study were in accordance with the ethical standards of the research ethics board of Faculty of Medicine and Dentistry, Palacký University Olomouc and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Fistulas occurring between the gastrointestinal and urinary systems are infrequent but are becoming more prevalent in current surgical and medical practice (9). These conditions encompass a diverse range of pathological entities, representing uncommon complications that can arise from both benign and malignant processes (10). Although very rare, a gastrovesical fistula can potentially develop in patients after undergoing endoscopic drainage for (peri) pancreatic fluid collections, as documented in our report. To our knowledge, this is the first case of such an outcome reported in the available literature.

The long-standing gold standard therapy for cases of severe NP had been open necrosectomy, although acknowledged for associated morbidity and mortality (7,11,12). With increasing reports highlighting the risks

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related to surgery, its utilization has been eventually limited to selected cases. The advent of ETD, coupled with the utilization of LAMSs and other endoprostheses, allowed a paradigm shift in the treatment of NP and WON (4,13,14). This approach combines the benefits of endoscopy and ultrasound imaging to precisely guide drainage. By utilizing an endoscope equipped with an ultrasound probe, the endoscopist can accurately identify the collection and safely access it through the stomach or duodenal wall, creating an internal connection between the necrotic cavity and the gastrointestinal tract with a stent. The technique offers improved outcomes with reduced AE rates, shorter hospital stays, and faster recovery times, solidifying its position as the method of choice (1,4,6).

A commonly employed strategy in the treatment of WON involves leaving transluminal plastic stents *in situ* following the removal of LAMS, aiming to enhance the drainage effects if required (5,15,16). Long-term transmural stenting proved to be a safe and effective modality (17). However, indwelling plastic stents-related AEs have been reported. Most of the literature highlights stent occlusion and dislocation. A case of small bowel obstruction due to stent migration was described (18). In addition, stents may erode into adjacent structures, especially the colon (17,19,20). Fortunately, these effects of transluminal plastic stents have been reported extremely seldom (17). We presented a unique case of gastrovesical fistula resulting from endoscopic drainage of WON.

In general, organ perforation secondary to stent placement can occur either as an early complication at stent insertion or as a late event due to stent migration or pressure necrosis (21). Theoretically, the formation of a gastrovesical fistula during or after endoscopic drainage of WON can occur due to several factors. One possible mechanism involves the erosion or injury to adjacent structures during the insertion and manipulation of endoscopic instruments within the necrotic cavity. The proximity of the necrotic collection to the bladder may also increase the likelihood of inadvertent tissue injury and subsequent fistula formation. Additionally, the necrotic tissue itself can contain proteolytic enzymes and inflammatory mediators that can further contribute to tissue breakdown and fistula formation (22). In our case, prolonged disease course, repeated procedures, and extended contact and pressure exerted by the plastic stents against surrounding structures, combined with inflammation and fibrosis, could all weaken tissue integrity and thus contribute to erosion. This resulted in gradual cavity extension and, eventually, fistula formation.

The presence of a bladder fistula following endoscopic drainage of WON may result in various clinical consequences and manifestations. The passage of gastric or duodenal contents into the bladder can cause pneumaturia, recurrent urinary tract infections, bladder irritation, and urinary symptoms such as urgency, frequency, and hematuria (9). Patients may also experience persistent abdominal pain and gastrointestinal symptoms such as nausea and vomiting. Elevated urinary amylase levels can serve as a marker for the presence of a pancreatico-vesical fistula. It is worth noting that our patient remained largely asymptomatic in this regard for most of the time. As soon as significant issues appeared, he was repeatedly referred to surgeons and urologists for possible radical surgery. However, such intentions were abandoned in all the instances by both specialties. There were no signs of acute abdomen, and any major procedure would likely be accompanied by additional, certainly major, complications. Judging by the development of this case, this approach eventually turned out to be the most favorable for the patient.

Management of a gastrovesical fistula requires recognition and an appropriate intervention. The treatment approach depends on the severity of the condition and the patient's overall health status. Initially, conservative measures may be employed. At this stage, the patient is usually treated in the general medicine ward or interdisciplinary intensive care units. Surgical consultation may be required to assess indication for the need of operative procedure, especially in cases of increased intraabdominal pressure unresponsive to conservative therapy, infected necrosis, or fulminant course of peritonitis. Gastrovesical fistula complicating the course of NP and/or related endoscopic drainage poses a distinct challenge. A surgical approach may be initially suggested, but it should be emphasized that invasive surgical interventions in severe inflammation, such as AP, are associated with a high number of AEs and increased morbidity and mortality (2,14).

Conclusions

Gastrovesical fistula can potentially arise as a complication following endoscopic drainage of WON through the gastrointestinal wall. Prompt recognition and a multidisciplinary approach involving surgeons, gastroenterologists, urologists, and radiologists are crucial in managing this event. Treatment involves conservative measures initially, but surgical repair may be required. Our patient was managed conservatively with the participation

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of several specialties. It emphasizes a rather tailored and non-invasive approach as a possible optimal management in these circumstances. To our knowledge, this is the first documented case of such an event. Given the increasing utilization of ETD for (peri)pancreatic collections, treating physicians should remain cautious and well-informed about all potential AEs, including those that are rare, associated with such procedures.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://acr.amegroups.com/article/view/10.21037/acr-24-18/rc

Peer Review File: Available at https://acr.amegroups.com/article/view/10.21037/acr-24-18/prf

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://acr.amegroups.com/article/view/10.21037/acr-24-18/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the research ethics board of Faculty of Medicine and Dentistry, Palacký University Olomouc and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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