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Case Report

Successful radiological embolization of a low output jejunal enterocutaneous fistula using a cyanoacrylate and lipiodol mixture

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ABSTRACT

Enterocutaneous fistulas (ECFs) are a significant complication of abdominal surgery which present a significant therapeutic challenge for clinicians. Definitive surgical treatment of these fistulas is not without significant risk, which both patients and clinicians would seek to avoid. We present a case of successful treatment of a low output jejunal ECF by percutaneous embolization using an ethiodized oil and n-butyl-2-cyanoacrylate glue mixture in a 40-year-old man who developed an ECF after inadvertent jejunostomy tube removal. This case demonstrates the technique and suitability of a widely available embolic agent for treatment of proximal small bowel ECFs. In addition, the case illustrates the potential of minimally invasive interventional radiological procedures in the management of ECFs, particularly in high-risk patients, as an alternative to surgery.

Introduction

Enterocutaneous fistulas (ECFs) are defined as an abnormal connection between the gastrointestinal tract and the skin. These most often form following abdominal surgery where there has been inadvertent intraoperative bowel injury or postoperative leak. Conservative management strategies involve wound care, treatment of infection, and nutritional support in order to promote spontaneous fistula closure. Despite this spontaneous closure rates are only in the order of 15%-25%, hence definitive fistula treatment is required in a large proportion of patients. Interventional radiology offers minimally invasive techniques that can mitigate the risks associated with further abdominal surgery, which can be significant in this cohort of patients. We present a case of a 40-year-old patient who developed a jejunal ECF following inadvertent jejunostomy feeding tube removal, after an initial admission and surgery for spontaneous oesophageal perforation. In our case we illustrate the successful fistula closure by percutaneous embolization using an ethiodized oil and n-butyl-2-cyanoacrylate glue mixture.

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Case report

A 40-year-old male was referred for interventional radiological management of a jejunal ECF following spontaneous oesophageal perforation. He initially presented with oesophageal perforation and Klebsiella empyema in November 2017. Significant past medical history included intravenous drug abuse and hepatitis C infection. He was then admitted to ITU after developing delirium, acute kidney injury and septic shock, leading to ITU admission. Relevant laboratory tests on admission included White cell count (WCC) $58.6 \times 10 \times 10\text{^}9/L$ and CRP 195 mg/dL. An initial CT scan with oral contrast demonstrated oesophageal perforation and right-sided pleural empyema. Emergency thoracotomy and laparotomy were performed to repair the ruptured oesophagus with oesophageal T-tube placement, placement of 2 surgical drains around the site of perforation, feeding jejunostomy, and draining gastrostomy. Surgical drains were removed following cessation of drain output.

A slow recovery followed in the initial postoperative period with persistent thoracic and abdominal infection, and significant wound dehiscence of the roof top incision that required supportive management including wound care, intravenous fluid replacement, and antibiotics/antifungals. In addition, the patient pulled out the feeding jejunostomy, which subsequently led over several weeks to the discharge of enteric contents from this jejunostomy site. CT findings (Fig. 1) confirmed ECF formation by demonstrating a jejunal loop subjacent to the abdominal wall at the site of previous feeding jejunostomy immediately adjacent an area of wound dehiscence, in addition to the passage of orally administered contrast into the wound externally via the ECF. Further CT excluded any abdominal collections. The upper gastrointestinal multidisciplinary meeting determined a nonsurgical approach regarding the ECF, aiming to achieve spontaneous closure after taking into account the patient's poor physical health, including poor wound healing and pleural infection, comorbidities, and complex social needs. A radiological guided insertion of a feeding gastrojejunostomy was determined not in the patient's best interest possible due to patient compliance at that time. The patient was discharged to the community with a plan for continued hospital review of the wound and ECF.

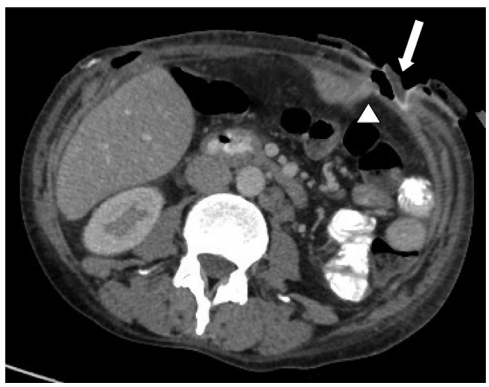

Fig. 1 – CT with portal venous phase and orally administered water-soluble contrast illustrating the formation of the enterocutaneous fistula. The image shows the jejunal loop subjacent to the abdominal wall (arrowhead) at the site of previous jejunostomy adjacent the area of wound dehiscence from the roof top incision wound, in addition to oral contrast seen within the wound externally (arrow).

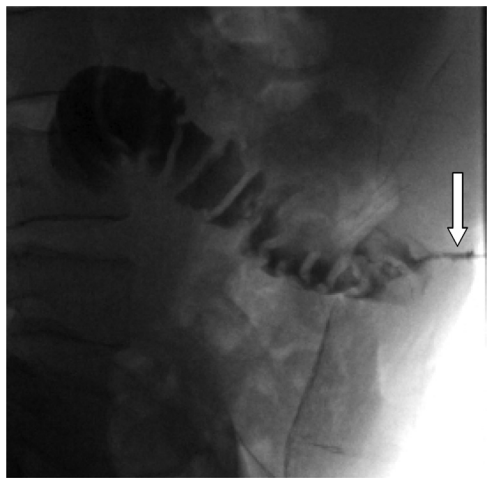

Fig. 2 – Lateral fluoroscopic view following catheter administration of water soluble contrast to delineate the jejunal enterocutaneous fistula (arrow), that demonstrates communication of the a 3 cm long fistula tract with the small bowel.

The patient was readmitted to our institution in August 2018 for management of the ECF, which was contributing significantly to his poor mental health and impaired activities of daily living. The patient was medically stable on admission with unremarkable laboratory investigations. Examination revealed an actively discharging fistula in the left anterior abdominal wall with an output of approximately 50-100 mL/day. A consensus decision for radiological treatment was determined after garnering opinions from both surgery and gastroenterology.

Embolization was performed on a dedicated interventional radiology procedural list by a Consultant Interventional Radiologist (equivalent to Attending Interventional Radiologist in North America). Anatomical delineation of the ECF was achieved through an initial fistulogram that demonstrated the communication of a 3 cm long fistula tract with the small bowel (Fig. 2). The embolization medium consisted of 0.5 mL of n-butyl-2-cyanoacrylate (Histoacryl; B Braun, Melsungen, Germany) and 2 mL ethiodized oil (Lipiodol; Guerbet Group), mixed using a standard intravenous 3-way tap. First the ECF was crossed from the cutaneous side with a standard Terumo nonvascular guidewire and Bernstein 5 Fr catheter. The wire and catheter were subsequently exchanged for a 90 cm Amplatz wire and 6 Fr Brite Tip Interventional Sheath, the latter positioned in the proximal/cutaneous end of the fistula (Fig. 3). With the Amplatz wire removed, the fistula was flushed with 200 mL 5% dextrose prior to emboliza-

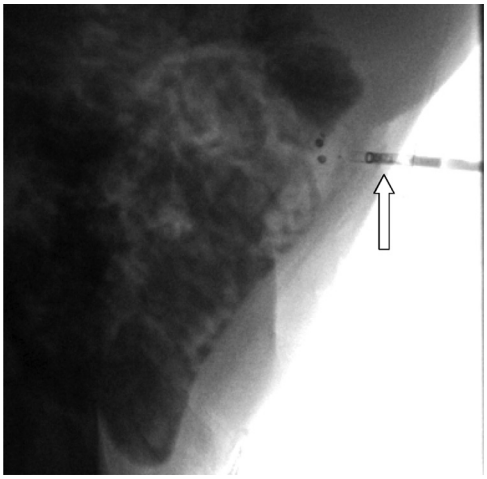

Fig. 3 – Lateral fluoroscopic view showing the 6 Fr Brite Tip sheath positioned in the proximal aspect of the fistulous tract (arrow) prior to embolization.

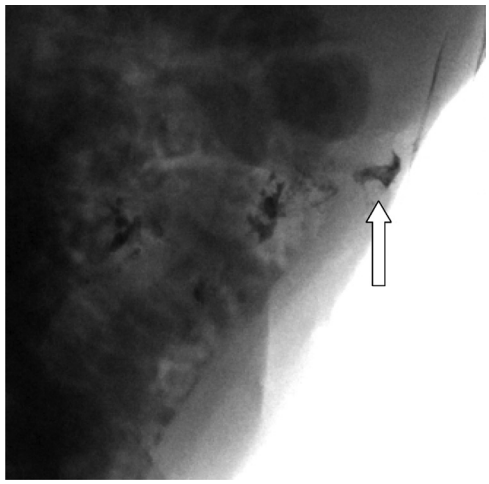

Fig. 5 – Lateral fluoroscopic view at the end of the procedure following catheter removal, demonstrating cyanoacrylate/lipiodol embolic agent within the enterocutaneous fistula tract (white arrow).

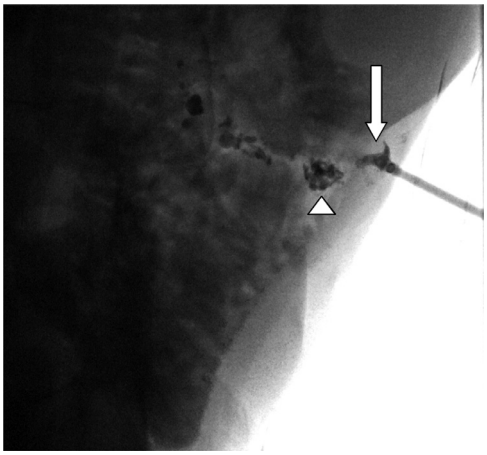

Fig. 4 – Lateral fluoroscopic view during manual injection of cyanoacrylate/lipiodol embolic agent. The contrast agent is seen in both the tract (arrow) and within the jejunum (arrowhead).

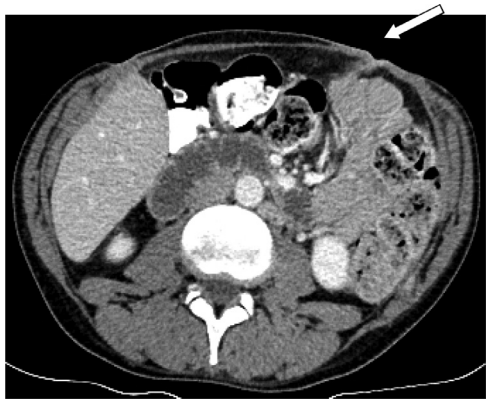

Fig. 6 – Postprocedure CT (8 days) with orally administered water-soluble contrast demonstrating closure of the fistula tract from the small bowel loop subjacent to the left-sided anterior abdominal wall. No contrast was identified within the remnant fistula tract or externally.

tion. The fistula tract was subsequently selectively embolized with the cyanoacrylate/ethiodized oil mixture (Fig. 4 and 5). The ethiodized oil component allowed fluoroscopic visualization of the delivery of the embolization material throughout the entire length of the ECF tract. There were no immediate complications. There was no evidence of fistula leakage postprocedure. Follow-up CT was performed on day 8 postembolization and identified that the ECF was no longer clearly demonstrated (Fig. 6). A small dimple formed in the left side of anterior abdominal wall that represented its previous position. The patient was reviewed on the ward 2 weeks following embolization, which confirmed cessation of fistula output and superficial wound granulation. The patient was discharged to the community 7 weeks following fistula embolization with no fistula output on discharge.

Discussion

ECFs are a significant complication of major abdominal surgery, which remain a significant therapeutic challenge despite modern advances. They are associated with significant rates of mortality, in addition to restriction to activities of daily living and mental health [1]. Therapeutic strategies are based around patient optimization, nutrition, and control of sepsis in order to promote spontaneous closure. While surgical procedures remain an option, they are not undertaken lightly and have a range of complications, which patients and clinicians would seek to avoid. Interventional radiology has the potential to offer minimally invasive techniques in the management of ECFs. Several case reports and series have described the successful closure of ECFs using a variety of embolization

techniques, including the use of n-butyl-2-cyanoacrylate/lipiodol mixtures; however, there is relative scarcity of reports of this technique for embolization of postoperative small bowel fistula. We present a case of successful treatment of a low output ECF by percutaneous embolization using n-butyl-2-cyanoacrylate glue in a 40-year-old patient.

ECFs are defined as abnormal connection between the gastrointestinal tract and the skin. They are classified according to the aetiology, anatomical level of involvement, and output. The majority are postsurgical (75%-85%) following inadvertent intraoperative bowel injury or postoperative anastomotic leak, for example in patients requiring surgery in the context of trauma, inflammatory bowel disease, or cancer [2]. The remaining "spontaneous" (15%-25%) fistulas occur in patients with inflammatory bowel disease, diverticular disease, appendicitis, cancer, radiotherapy, and ischaemia. Fistula output can be further described in 3 groups. A high-output fistula is defined as fistula output exceeding 500 mL/24 hours, moderate-output 200-500 mL/24 hours and low-output < 200 mL/24 hours [2,3].

Fistulas may close spontaneously in the long-term, with closure rate often quoted around 15%-25% [2]. This rate may vary depending on a variety of factors such as underlying the aetiology, fistula complexity, length, and output [4,5]. The anatomical level of involvement is also an important factor. While ECFs may occur at any level, distal fistulae are associated with poorer spontaneous closure. A recent study of 154 patients demonstrated a spontaneous closure rate of 74.8% of patients with ECFs proximal to the duodenal-jejunal flexure, compared to the 35.4% in distal fistulas [6]. The most favorable fistula profile for spontaneous closure are those with low output postoperative fistulas without infective or noninfective complications [4].

Despite the significant advances in surgical and nonsurgical management, ECFs are associated with a significant burden of mortality ranging between 6%-33% [7]. This is often higher in patients with sepsis and nutritional deficits [6,8]. Hence management strategies are centered on patient optimization to promote spontaneous closure focusing on management of sepsis, nutrition, and anatomical considerations [3,9]. Definitive repair of the ECF should be considered if spontaneous closure fails to occur by 12 weeks. Surgical closure rates, involving resection of the involved segment and anastomoses, ranges between 75% and 85% [10] but are associated with complications including mortality which is incurred at a rate of up to 4.8% [11].

Minimally invasive techniques offered by interventional radiology have the potential to obviate the need for further surgical intervention and avoid postoperative complications. Cyanoacrylate glues have been used off-label for percutaneous embolization for some time. These glues form strong adhesive bonds to soft tissues formed upon contact with bodily fluids [12]. It remains a popular embolic agent due to its ubiquity, low cost, and familiarity with interventionists. A common practice involves the combination with ethiodized oil to form a 2-part embolic agent. This practice not only opacifies the embolic agent but prolongs the polymerization time thus facilitating its administration [13].

While much has been published regarding the use of n-butyl-2-cyanoacrylate glue embolization for ECFs, there have been few published reports of this technique for embolization of postoperative fistula involving the jejunum and ileum [14]. Mauri et al describes the successful closure of 3 ECFs on first attempt including an jejunal fistula, which developed shortly after subtotal colectomy, using a mix of glubran-2 (a variant cyanoacrylate glue with longer polymerization time) and lipiodol [15]. In a separate case series of 18 patients, the same author reported successful closure of a variety of postoperative ECFs using the same method [16]. One of the successful closures was in a patient with a jejunal fistula, however this required a total of 3 glue injections and required 25 days for healing.

Other types of fistula have been successfully closed with this technique. Cambj–Sapunar et al reported the successful embolization of 7 ECFs in 6 patients, although the fistulas were all involved the colon rather than small bowel [17]. Bae et al reported the closure of 11 enteric and biliary fistulas with cyanoacrylate glue injection (Histoacryl) with successful closure after 1 or 2 treatment sessions without complication [18]. The successful use of other embolic materials has also been published including gelfoam (Pfizer Pharmaceuticals, New York, NY) [19] and ethylene-vinyl alcohol copolymer (Onyx 34; Micro Therapeutics, Inc., Irvine, CA) [20]. These alternative techniques provide further options for embolization of ECFs. The choice of embolic agent; however, will be determined by factors including fistula type and complexity, availability of embolic agent, and cost. The familiarity and ubiquity of n-butyl-2-cyanoacrylate glues ensure that it remains popular embolic agent for many cases.

In our case we presented the successful closure of a low output postoperative jejunal ECF. The patient had significant comorbidities contributing to an overall unfavorable profile for spontaneous ECF closure including poor nutrition, infrequent wound care, short fistula length and recurrent thoracic infection, and pleural collections. The technique provided successful following a single application of cyanoacrylate glue in a fistula that had failed to spontaneously close in 9 months. The postprocedure result is similar to the experience of the few other published cases regarding embolized jejunal ECFs. This case provides further evidence of the utility of this technique in small bowel ECFe in which the spontaneous closure has not occurred.

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