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Recurrent Type 1 Enterocutaneous Fistula and Granulomatous Gastritis: A Case Report

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 34-year-old
Final Diagnosis: Enterocutaneous fistula
Symptoms: Abdominal pain • fistula
Medication: —
Clinical Procedure: Distal subtotal gastrectomy
Specialty: Surgery

Objective: Unusual clinical course**Background:** Enterocutaneous fistula is an abnormal communication between the gastrointestinal tract and skin. One-third of enterocutaneous fistulas disappear spontaneously, but the rest of them require surgical treatment.**Case Report:** We describe the case of a 34-year-old woman with enterocutaneous fistula that she had had for year. She had previously undergone 2 unsuccessful operations, and the fistula recurred twice. Distal subtotal gastrectomy and Billroth 2 reconstruction were performed. In the pathological examination, the distal gastrectomy specimen revealed foci of ulceration. Well-circumscribed non-necrotizing granulomas were occasionally encountered beneath the ulcers.**Conclusions:** Failure of treatment in recurrent fistula management has a variety of reasons. Our clinical experience shows that granulomatous gastritis can be a complicating factor in treatment of recurrent enterocutaneous fistula.**Keywords:** Celiac Disease • Gastrectomy • Granuloma, Giant Cell • Intestinal Fistula**Full-text PDF:** <https://www.amjcaserep.com/abstract/index/idArt/928532>

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Background

Enterocutaneous fistulas (ECF) are aberrant connections between the gastrointestinal tract and skin. They are classified according to their daily output, etiology, and source [1]. If the daily output is more significant than 500 ml, it is classified as a high-output fistula. If the daily output is less than 200 ml, it is classified as a low-output fistula. If the daily output is 200-500 ml, it is classified as a moderate output fistula. Most (75-85%) ECFs are iatrogenic and the rest occur spontaneously. The most common causes are intra-abdominal surgery, abdominal trauma, and inflammatory bowel diseases. There are 4 types of enterocutaneous fistulas, classified according to their anatomic localization: type 1 (esophageal, gastroduodenal), type 2 (small bowel), type 3 (colon), and type 4 (enteroatmospheric). About 20-30% of enterocutaneous fistulas heal spontaneously [2].

Case Report

A 34-year-old-woman with celiac disease presented at the General Surgery Department with leakage of gastrointestinal fluid out of her upper abdominal wall. Her medical history revealed the problem started 10 years before, and she had undergone 2 unsuccessful fistula takedown operations. During her inpatient evaluation, the daily output of the fistula was approximately 300 ml per day. Her body mass index was 20.8, and prealbumin level was 9.6 mg/dl. An abdominal computer tomography scan showed that the fistula tract was anterior to the left lobe of the liver. It was 9 mm in thickness and 6 cm long. The fistula tract was filled with air and oral contrast material. The communication of the fistula tract to any organ was not determined (Figure 1).

Conventional fistulography was performed to demonstrate the communication of the fistula tract. The fistula tract was between the first part of the duodenum and skin (Figure 2).

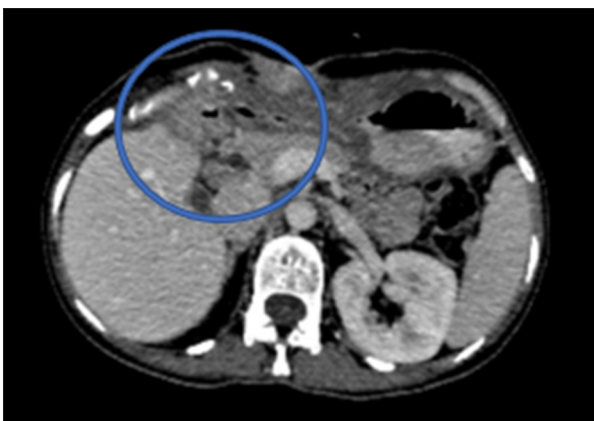


Figure 1. The circle shows the fistula tract and subcutaneous abscess cavity filled with air and oral contrast material.

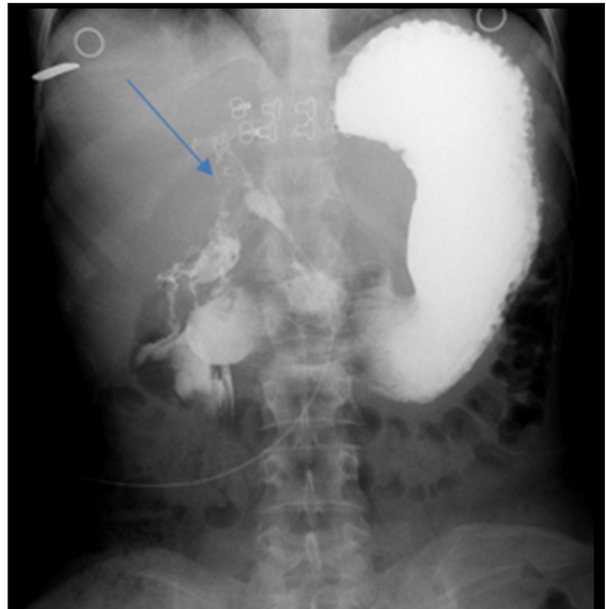


Figure 2. The arrow shows that the fistula tract originates from the first part of the duodenum.



Figure 3. The arrows show the fistula openings.

Esophagogastroduodenoscopy was performed, showing the pylorus was destroyed and the bulbus lumen was remarkably shrunken. The fistula opening was not seen during the examination.

After the preoperative evaluation, the decision was made to perform surgery. First, methylene blue dye was injected into the 3 fistula openings, and a stylus was used to see the pathway of the fistula tract (Figure 3). A midline incision was made,

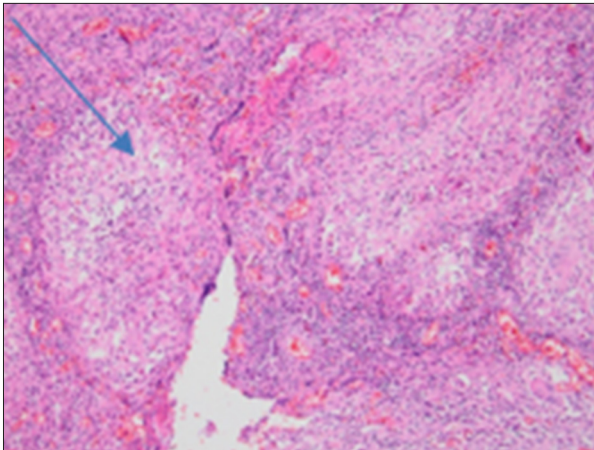


Figure 4. The arrow shows well-circumscribed granulomas were occasionally present beneath the ulcers.

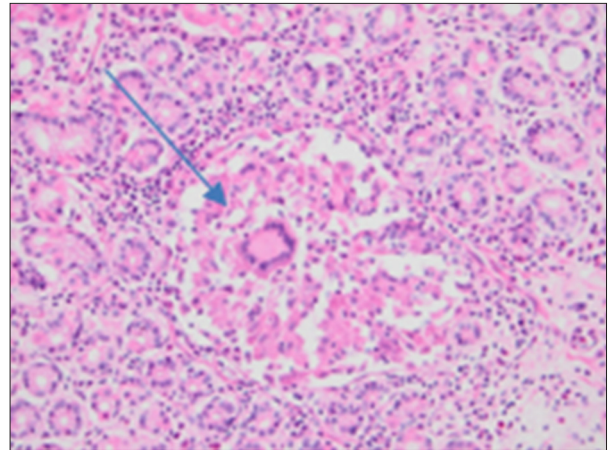


Figure 5. The arrow shows non-necrotizing granulom in a stomach sample.

and it was seen that one of the fistula tracts was opening into the pylorus, and the other tractus was opening into the first part of the duodenum. Because of the destruction of the natural anatomy of the pylorus and duodenum, distal subtotal gastrectomy and Billroth 2 reconstruction were performed, then the fistula tracts were de-epithelized and curetted. The post-operative course was unremarkable, and she was discharged on the fifth postoperative day. At the 6-month follow-up, she reported no problems.

In the pathological examination, the distal gastrectomy specimen revealed foci of ulceration. Well-circumscribed granulomas were occasionally encountered beneath the ulcers (Figure 4 H&E 10 \times). Granulomas, characterized by epithelioid histiocytes, and multinucleated giant cells surrounded by lymphoid and plasma cells were also present superficially in the lamina propria between the glands. The granulomas found in stomach samples were non-necrotizing (Figure 5 H&E 20 \times). In 1 lymph node dissected from perigastric fat tissue on the specimen, granuloma with central necrosis was also detected (Figure 6 H&E 10 \times). The lymph node was also evaluated with special stains for possible mycobacterial and fungal etiology, which were negative. Still, it was recommended that the patient be investigated clinically for a systemic granulomatous disorder.

Discussion

In this case report, we present a patient with recurrent type 1 ECF who had previously undergone 2 unsuccessful operations. The etiology of ECF is multifactorial. Almost 80% of ECFs occur following surgical operations, trauma, and inflammatory bowel disease. The present patient had no intra-abdominal operations, trauma history, or known inflammatory bowel disease. Nonsurgical etiology, presence of recurrent and multiple fistulas, delay getting to a tertiary hospital, and gastric

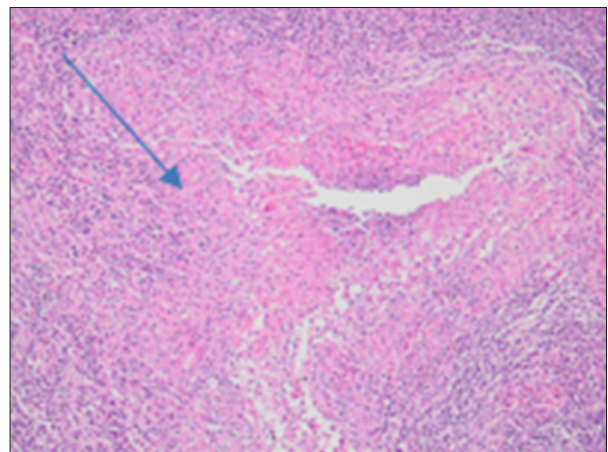


Figure 6. The arrow shows lymph node specimen granuloma with central necrosis.

origin of the fistula are unfavorable factors predicting nonoperative fistula closure, as in this case [1].

According to the literature, the recurrence rate of surgically treated enterocutaneous fistulas is nearly 20%. The surgical repair technique used is significantly associated with recurrence. A prospective study found that patients with a wedge repair or oversewing of an ECF had a recurrence rate of 32.7%, and patients with resection or anastomosis revision had a recurrence rate of 18.4% [3]. In the present patient, we performed resection, but the 2 previous operations were the only repair of the fistula tract.

In enterocutaneous fistula management, 4 parameters determine the morbidity and mortality risk of these patients: fluid-electrolyte balance, sepsis, nutritional status, and skin/wound care [4]. Our patient had border-line malnutrition, and her body mass index was relatively low.

The reason for the failure of previous treatments could also be the granulomatous gastritis in the patient. In the literature, there are no studies showing a correlation between these 2 entities. The patient was diagnosed with celiac disease in another hospital, so the appropriate tests were not performed in our hospital. We could not contact the patient because of technical reasons, so we could not perform a detailed diagnostic work-up for granulomatous gastritis or inflammatory bowel diseases.

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Conclusions

Recurrent enterocutaneous fistulas require detailed preoperative evaluation, sufficient nutritional support, adequate fluid-electrolyte balance, and sepsis management. The timing of surgery should be decided after adequate resuscitation, and resection should be the preferred surgical treatment. Our clinical experience shows that granulomatous gastritis can be a complicating factor in treatment of recurrent enterocutaneous fistula.