

3. Horii T, Ohta T, Mori T, Sakai M, Hori N, Yamaguchi K, et al. Ciliated hepatic foregut cyst. A report of one case and a review of the literature. *Hepatol Res.* 2003;26:243-8.
4. Wilson JM, Groeschl R, George B, Turaga KK, Patel PJ, Saeian K, et al. Ciliated hepatic cyst leading to squamous cell carcinoma of the liver. A case report and review of the literature. *Int J Surg Case Rep.* 2013;4:972-5.
5. Sharma S, Dean AG, Corn A, Kohli V, Wright HI, Sebastian A, et al. Ciliated hepatic foregut cyst: an increasingly diagnosed condition. *Hepatobiliary Pancreat Dis Int.* 2008;7:581-9.
6. Bishop KC, Perrino CM, Ruzinova MB, Brunt EM. Ciliated hepatic foregut cyst: a report of 6 cases and a review of the English literature. *Diagn Pathol.* 2015;10:81-6.
7. Harty MP, Hebra A, Ruchelli ED, Schnauffer L. Ciliated hepatic foregut cyst causing portal hypertension in an adolescent. *Am J Roentgenol.* 1998;170:688-90.
8. Fernández-Aceñero MJ, Corral JL, Manzarbeitia F. Ciliated hepatic foregut cyst: two further cases with an immunohistochemical analysis. *Hepatogastroenterology.* 2012;59:1260-2.
9. Goodman MD, Mak GZ, Reynolds JP, Tevar AD, Pritts TA. Laparoscopic excision of a ciliated hepatic foregut cyst. *JSLs.* 2009;13:96-100.
10. Borner MM. Neoadjuvant chemotherapy for unresectable liver metastases of colorectal cancer-too good to be true? *Ann Oncol.* 1999;10:623-6.

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Enteral Nutrition in Crohn's Disease With a High Output Enteroatmospheric Fistula^{☆,☆☆}



Nutrición enteral en enfermedad de Crohn con fístula enteroatmosférica de alto débito

Enteroatmospheric fistulae (EAF) are a subgroup of enterocutaneous fistulae (ECF) in patients with laparostomy or open abdomen. They are characterized by being superficial, with a high volume of discharge and surrounded by viscera or granulation tissue.¹⁻⁴ These factors can lead to a situation of metabolic and water-electrolyte imbalance, sepsis and severe malnutrition.

The conservative approach in functional short bowel syndrome associated with a complete jejunal fistula, using the reinfusion of the proximal fistula discharge through the distal jejunostomy, provides good results. In the physiological approach that we propose, we combine the reintroduction of the discharge with artificial enteral nutrition, which achieves improved nutritional state, a reduction in comorbidities and greater recovery of the intestinal mucosa, thereby facilitating the surgical closure of the EAF.

We present the case of a 19-year-old male with penetrating/stenosing Crohn's disease (CD), diagnosed 11 years earlier, with perianal involvement and ileocecal disease, which had had a terminal colostomy due to severe stenosis of the sigma 3 years earlier. He had presented with pancolitis and steroid-dependent extensive ileitis, refractory to various lines of biological treatment and currently treated

with ustekinumab, with favorable clinical, radiological and endoscopic response, but stenotic changes persisted at the ileal level. The patient was hospitalized due to abdominal pain secondary to ileal stenosis and reactivation of his CD. During hospitalization, he presented symptoms of bowel obstruction that required emergency surgery, involving ileocecal resection after meticulous adhesiolysis. On the 10th day post-op, the patient was re-operated due to bowel leak, observing a catastrophic abdomen with involvement of multiple loops, so we opted for damage-control surgery and laparostomy assisted with negative pressure therapy for later surgical revision or a "second look".

In the postoperative period, a complete fistula persistence was observed with high discharge with the afferent and efferent loops in the proximal jejunum, which resulted in a situation of functional short bowel syndrome. Initially, parenteral nutritional support was initiated, but worsened hepatic function was observed, with cytotoxicity of multifactorial etiology (fasting, infection, total PN, medication).

Given the situation of important malnutrition (41 kg, BMI: 15.74 kg/m²) and comorbidity associated with the use of parenteral nutrition, we considered optimizing the enteral nutritional support by means of a physiological model,

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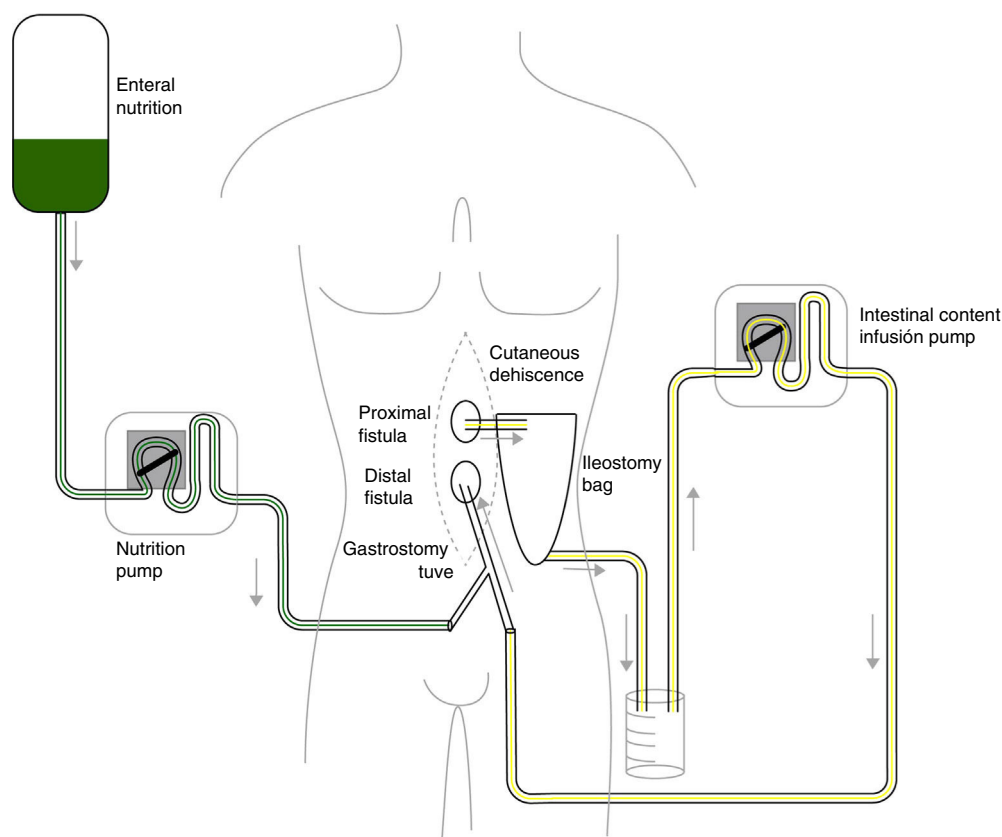


Fig. 1 – Physiological model for collecting and reinfusing the jejunal fistula discharge associated with enteral nutrition.

using continuous infusion of the nutritional product and the discharge collected from the fistulous orifice through 2 simultaneous pumps connected in a Y (Fig. 1). First of all, in order to collect the proximal discharge in a laparostomy with vacuum-assisted closure, the fistulous orifices were isolated with moldable discs, paste and an ileostomy bag connected to a closed drainage system. For the administration of enteral nutrition (EN) and the discharge from the proximal fistulous orifice, we utilized a 24 F gastrostomy catheter in the distal fistulous orifice, through the ileostomy bag. In order to minimize leaks, we used an adapted cone with adhesive. We consider it recommendable to fill the balloon of the gastrostomy catheter above the indicated value, which in our case was 2 cc, in order to guarantee a tight seal (requiring adaptation to the condition and diameter of the jejunum) (Fig. 2A–D). Six weeks after treatment, we observed normalization of the hepatobiliary function and improved nutritional parameters (52 kg, BMI: 19.42 kg/m²). Four months later, an adequate nutritional state was reached to schedule surgery, which involved resection of the fistulized jejunal loop, manual end-to-end primary anastomosis and reconstruction of the abdominal wall with biological mesh and animal collagen. Currently, the patient is being followed up in the outpatient clinic and has shown positive clinical and nutritional evolution with an ordinary diet.

Nutritional therapy is an essential part of the management of ECF due to the rapid and inevitable protein catabolism.^{5,6} Unfortunately, there are no randomized controlled clinical trials about their overall management, which limits the ability to establish specific nutritional support strategies. However, it is clear that the treatment of these patients requires specialized surgeons backed by a multidisciplinary team (MDT) with experience in the treatment of complex cases.⁷ In Spain, the implementation of these multidisciplinary units has been slow, and ours is the first in our region.

In patients with EAF, we are faced with the anatomical difficulties secondary to an open abdomen.³ This continuous infusion system of the proximal fistula discharge by means of a gastrostomy catheter resolves the problems derived from the poor fixation of other catheters to the efferent loop. Our experience indicates that small variations may be necessary in the placement of these devices, depending on the anatomical situation of the fistula. Furthermore, periodical revisions may be needed to avoid complications secondary to decubitus caused by the balloon.

This continuous reinfusion leads to maximal optimization of EN support and provides the ability to decrease or discontinue parenteral nutrition (PN), which has classically been more widely used.⁸ This results in fewer complications secondary to prolonged PN, such as improved hepatobiliary

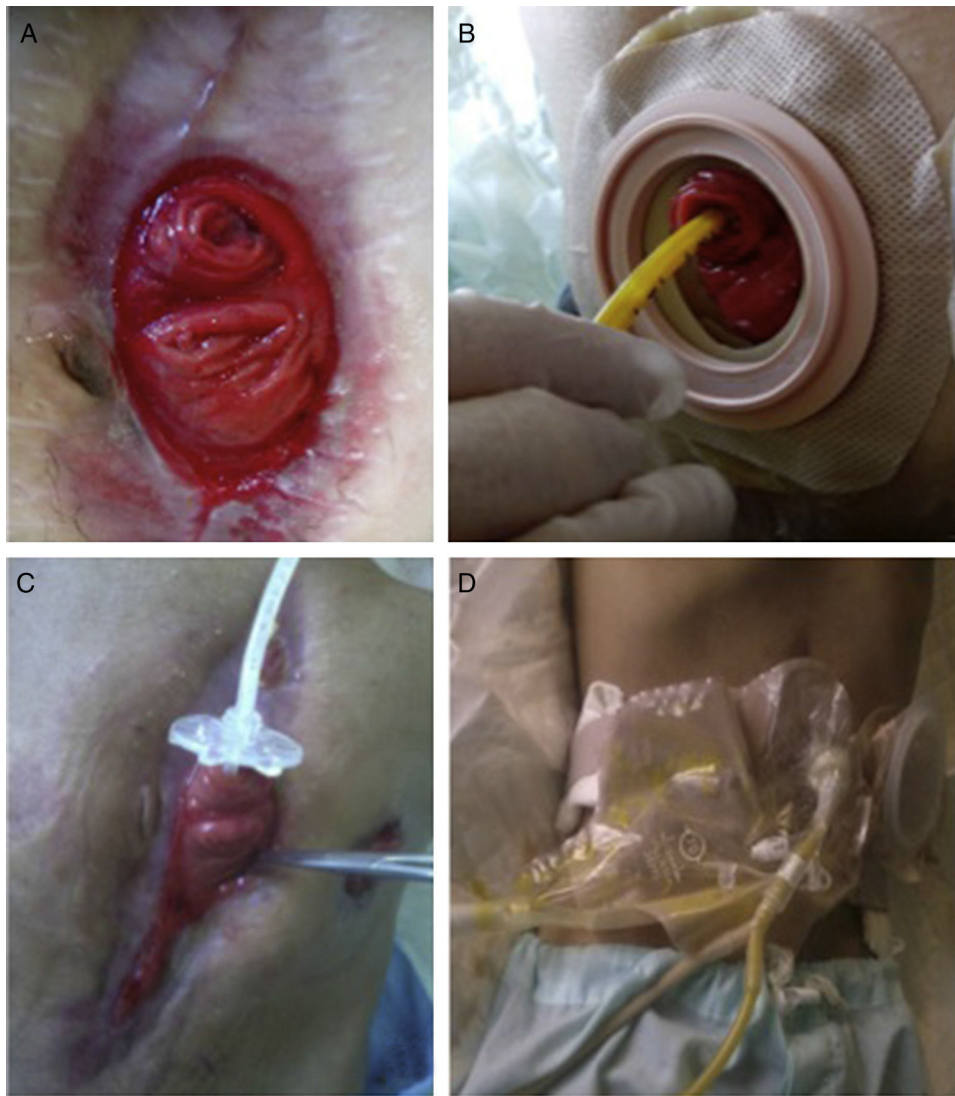


Fig. 2 - (A) Enterocutaneous fistula; (B) distal fistulous orifice canalized with a gastrostomy tube; (C) proximal fistulous orifice indicated with forceps; and (D) collection and infusion system using a physiological model.

function and the lower risk of sepsis associated with central venous catheters. It also facilitates the recovery of trophism, achieving the main objective of providing high levels of calories and nutrients.^{9,10} As a whole, these factors optimize the nutritional state and ensure better results in the definitive surgical treatment for closing the jejunal fistula.

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Conflict of Interests

The authors have no conflicts of interests to declare.

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REFERENCES

1. Connolly PT, Teubner A, Lees PN. Outcome of reconstructive surgery for intestinal fistula in the open abdomen. *Ann Surg.* 2008;247:440-4.
2. Davis KG, Johnson EK. Controversies in the care of the enterocutaneous fistula. *Surg Clin N Am.* 2013;93:231-50.

3. Waisntein DE, Delgado M, Sisco P. Tratamiento de 50 casos de fístulas enteroatmosféricas. In: 82 Congreso Argentino de Cirugía; 2011.
4. Waisntein DE, Irigoyen M, Beninka E. Fístulas enterocutáneas. In: Capítulo III-255 Enciclopedia Cirugía Digestiva F. Galindo et al.. 2014. p. 1-29.
5. Rodríguez Cano AM. Terapia nutricia en fístula enterocutánea; de la base fisiológica al tratamiento individualizado. *Nutr Hosp.* 2014;29:37-49.
6. Vallés-Gamboa M, Lahaba-Liquí N, Castillo-Gutiérrez O. Fístulas enterocutáneas de intestino delgado. *Manejo terapéutico.* 2002;3.
7. Mawdsley JE, Irving PM, Makins RJ, Rampton DS. Optimizing quality of outpatient care for patients with inflammatory bowel disease: the importance of specialist clinics. *Eur J Gastroenterol Hepatol.* 2006;18:249-53.
8. Pironi L, Arends J, Baxter J, Bozzetti F, Peláez RB, Cuerda C, et al., Home Artificial Nutrition & Chronic Intestinal Failure; Acute Intestinal Failure Special Interest Groups of F ESPEN. ESPEN endorsed recommendations. Definition and classification of intestinal failure in adults. *Clin Nutr.* 2015;34:171-80.
9. Dudrick SJ, Panait L. Metabolic consequences of patients with gastrointestinal fistulas. *Eur J Trauma Emerg Surg.* 2011;37:215-25.
10. Pflug AM, Utiyama EM, Fontes B, Faro M, Rasslan S. Continuous reinfusion of succus entericus associated with fistuloclysis in the management of a complex jejunal fistula on the abdominal wall. *Int J Surg Case Rep.* 2013;4:716-8.

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