

Combined treatment of enterocutaneous fistula with laser diode and embolization

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ABSTRACT

Introduction. Enterocutaneous fistula treatment in patients undergoing multiple surgeries is complex and requires creative solutions. We present the case of an enterocutaneous fistula managed with laser diode and cyanoacrylates.

Clinical case. 15-year-old patient, diagnosed with ulcerative colitis at 12 years of age, undergoing full colectomy with urgent ileostomy as a result of a flare-up refractory to medical treatment. Five months later, an ileoanal pull-through with pouch was carried out, leaving a protection ileostomy in place. However, postoperative evolution was poor, with pelvic infection, so two further urgent open surgeries were required for lavage and hemostasis purposes.

Six months later, anastomotic stricture was noted. It was healed following various pneumatic dilations under ultrasound vision and at-home dilations using Hegar dilators. One year following this, ileostomy was closed, but one month later, abdominal distension occurred. It was associated with a fistula in the abdominal midline, which could be endoscopically guided, with its origin being located at the ileoanal anastomosis. Laser diode sessions were applied for treatment purposes, with partial improvement, but still with gas emission. One year later, embolization was performed by placing platinum coils and lipiodol-diluted cyanoacrylates, and clinical signs disappeared. 17 months following this surgery, the patient has no symptoms, with full day and night fecal continence and 3 daily stools, and the fistula is completely closed.

Conclusion. Combined treatment with laser diode and platinum coil and cyanoacrylate embolization proves effective in the management of enterocutaneous fistula, with low morbidity.

KEY WORDS: Enterocutaneous fistula; Laser; Cyanoacrylates.

TRATAMIENTO DE FÍSTULA ENTEROCUTÁNEA COMBINANDO LÁSER DIODO Y EMBOLIZACIÓN

RESUMEN

Introducción. El tratamiento de las fístulas enterocutáneas establecidas en pacientes multioperados es complejo y requiere de soluciones creativas. Presentamos la resolución de un caso con láser diodo y con cianoacrilatos.

Caso clínico. Paciente de 15 años, diagnosticado a los 12 de colitis ulcerosa y sometido a pancolectomía con ileostomía urgente por brote refractario al tratamiento médico. Cinco meses después se realizó un descenso ileoanal con reservorio dejando ileostomía de protección, pero presentó un posoperatorio tórpido con infección pélvica, precisando dos nuevas laparotomías urgentes, para lavado y hemostasia.

Seis meses después, se evidenció una estenosis de la anastomosis que se resolvió tras varias dilataciones, neumáticas bajo visión endoscópica y domiciliarias con tallos de Hegar. Un año después se cerró la ileostomía, presentando al mes una distensión abdominal y aparición de fistula en línea media abdominal, que pudo tutorizarse endoscópicamente, observando su origen en la anastomosis ileoanal. Se trató mediante sesiones de láser diodo, con mejoría parcial, aunque persistía la salida de gases. Un año después se embolizó implantando espirales de platino y cianoacrilatos diluidos con lipiodol, resolviéndose totalmente el cuadro. Transcurridos 17 meses de esta última intervención, el paciente se encuentra asintomático, con continencia fecal total diurna y nocturna, tres deposiciones al día y la fístula está totalmente cerrada.

Conclusión. El tratamiento con láser diodo, combinado con embolización con espirales de platino y cianoacrilatos, puede ser un tratamiento eficaz con baja morbilidad de las fístulas enterocutáneas establecidas.

PALABRAS CLAVE: Fístula enterocutánea; Láser; Cianoacrilatos.

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INTRODUCTION

Enteric fistula is a severe complication following abdominal surgery. It is difficult to treat, and it often makes hospital stay significantly longer and increases anxiety as a result of feeling surgery was not completely satisfac-

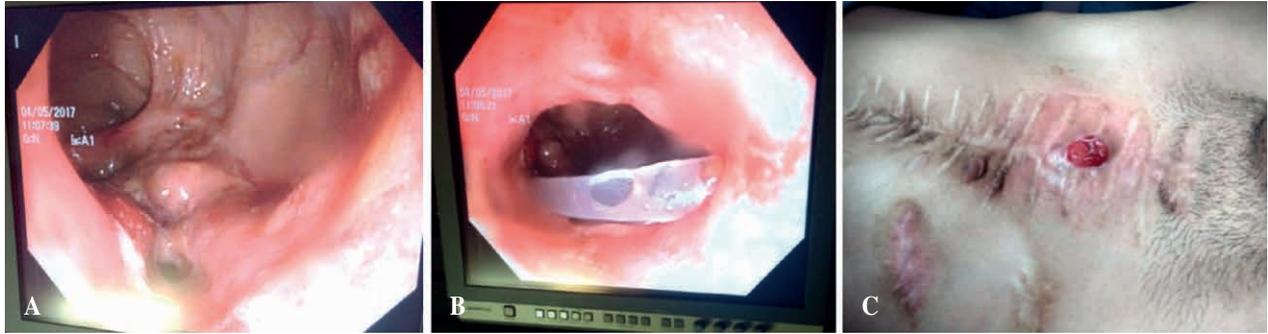


Figure 1. Endoscopic guidance of the fistula, with its tract being located from the abdominal midline to the ileoanal anastomosis. A) Exit orifice of the fistula at the level of the anastomosis. B) Fistula guidance from the abdomen using a probe. C) External look of the fistula.

tory. Therefore, it has a considerably negative impact on patients' quality of life⁽¹⁾.

In children, enteric fistula treatment is variable and depends on many factors, such as duration, flow, location, patient pathology, and number of previous surgeries. Sometimes, especially in younger children, enteric fistula can be spontaneously healed with conservative treatment. Various medical treatments such as somatostatin or some of its analogues, such as octreotide, have proved beneficial as they reduce pancreatic enzyme secretion and intestinal motility, thus accelerating fistula closure. However, they do have cardiovascular and digestive side-effects, and the need for subcutaneous or intravenous administration is to be considered, since it impairs long-term use⁽²⁾. If the fistula is large and associated with partial abdominal cavity removal, it typically becomes chronic, while slowly closing the abdominal wall, thus adopting the shape of a "neostoma".

Re-intervention, especially deferred re-intervention, is sometimes required to completely solve this. However, this surgery is not risk-free as it can cause new perforations, and even intestinal resection may prove necessary. The situation is even more complex if the fistula is located in an extraperitoneal region, such as the area caudal to peritoneal reflection. This typically occurs following ileoanal pull-through, which is sometimes required in children with Hirschsprung's disease or colon inflammatory disease, such as ulcerative colitis. Surgical management in these cases is highly complex, since the surgical approach is difficult and it often requires protection ileostomy. This is particularly hard to understand for patients and their families, with the added risk of causing patient incontinence if the dentate line is damaged and/or adjacent genitourinary structures are injured.

Therefore, these complex patients require a cross-disciplinary treatment with effective but less invasive alternatives. Sometimes, solutions used in other pathologies or specialties may prove useful, such as interventional radiology, which is able to obstruct newly formed

tracts^(3,4), or laser diode, which is used in fistulas at other locations⁽⁵⁾.

In the literature, there are few recurrences of this pathology in the pediatric population, with cases always being individual ones^(6,7). This is the case of a pediatric patient with poor postoperative evolution following ileoanal pull-through who developed enterocutaneous fistula. The fistula was conservatively healed thanks to laser fulguration and cyanoacrylate and platinum coil embolization.

CLINICAL CASE

We present the case of a 16-year-old patient diagnosed with ulcerative colitis refractory to medical treatment at 12 years of age who underwent emergency full colectomy with protection ileostomy.

Five months later, an ileoanal pull-through with J-pouch was carried out, leaving a protection ileostomy in place. However, postoperative evolution was poor, with pelvic infection, so two further urgent open surgeries were required for lavage and hemostasis purposes, while leaving various drainages in place at that level.

Six months later, anastomotic stricture was noted. It was healed following various pneumatic dilations under ultrasound vision and several poorly-tolerated at-home dilations using Hegar dilators. One year following this, the patient requested the ileostomy was closed, which was attempted in spite of the risk of complications. In the pre-closure opacification, a small slot compatible with residual fistula was found at the level of the anastomosis, but it could not be confirmed. One month following ileostomy closure, the patient had abdominal distension, and 48 hours later, a fistula was observed in the abdominal midline, through which feces with the same look as those from the previous ileostomy were expelled. The fistula could be endoscopically guided and visualized under general anesthesia, with its origin being located at the ileoanal anastomosis (Fig. 1).



Figure 2. Radiological image of cyanoacrylate and platinum coil embolization.

Over the next year, the patient was treated with 3 laser diode sessions with radial firing fiber (power: 12 watts; wavelength: 1,470 nanometers), with the probe being introduced through the fistula until the perineum. They were performed at the operating room, under general anesthesia. Improvement was partial, with persisting malodorous gases, which prevented the ostomy bag from being removed. Once, the perianal area was burnt, which required high analgesic doses, so decision was made to discontinue treatment. On the other hand, anal feces became more frequent and well-formed.

One year following the first laser session, the fistula was embolized with the help of the interventional radiology department. Under general anesthesia, platinum coils and lipiodol-diluted cyanoacrylates were placed along the whole fistulous tract, and clinical signs disappeared (Fig. 2). The ostomy bag was removed, and a gauze was placed in the fistulous area, which was fully closed. Occasionally, he expelled a platinum coil along with a minimal exudate, so decision was made to repeat the procedure two months later. Since then, 17 months following the last embolization, the patient has had no symptoms, with total day and night fecal continence and 3 daily stools. The fistula is now closed. The patient is currently 17 years old, he plays sports, and he leads a completely normal life for his age.

DISCUSSION

Enterocutaneous fistula typically occurs following a complex abdominal surgery and sparks a cascade of events, such as infection, obstruction, and poor general condition, which can significantly increase patient morbidity and even mortality^(1,8). This is the case of an adolescent with intestinal inflammatory disease – ulcerative colitis – who had previously undergone immunosuppressive treatments to

slow down activity and a complex ileoanal pull-through surgery with pouch. Perineal surgical time was complex and long, which favored the occurrence of a pelvic abscess during the immediate postoperative period. It is unclear whether the fistula –and therefore the abscess– was caused by partial anastomotic dehiscence or ischemia secondary to tension of the pulled-through segment, or if the abscess was accountable for the poor anastomotic healing, and therefore, the fistula. However, in this situation, the patient can be offered limited surgical options with a guarantee of success. He improved thanks to the stoma, but anastomotic stricture and the need for dilation had a negative impact on his self-esteem. As a result of this, the patient lost weight and stopped leading a normal life.

Once the stricture was considered to be over, and given the patient's insistence, the ileostomy was closed, which revealed the presence of a more chronic and established fistula than expected. The proximity of the anastomosis to the dentate line deterred us from performing a new pull-through owing to the risk of causing permanent fecal incontinence or damaging genitourinary structures such as the ductus deferens or the seminal vesicles in a fibrotic or “frozen pelvis” context.

Laser diode results have proved variable, especially in adults⁽⁵⁾, and they have been mainly described in the management of perianal fistula, not postoperative enterocutaneous fistula. The idea of burning the tract to prevent reproduction sounded compelling in order to occlude the fistula. It was attempted in 3 sessions, with a clear improvement, since it significantly reduced flow through the fistula. However, as a result of gravity, gas still had a more favorable way out through the fistula than through the anus. Therefore, and also because the laser once burnt the perineum –which required high analgesic doses–, tract embolization with cyanoacrylate and platinum coil placement was considered, which made symptoms fully disappear. These substances have been used to occlude other fistulous tracts secondary to surgical complications, but the literature only provides with isolated cases, and virtually none in pediatric patients⁽⁷⁾.

In conclusion, combined treatment with laser diode and platinum coil and cyanoacrylate embolization proved highly effective and very little morbid in this patient. Therefore, it should be considered as one of the therapeutic options available in case of well-established enterocutaneous fistulas which are difficult to manage surgically.

REFERENCES

1. Falconi M, Pederzoli P. The relevance of gastrointestinal fistulae in clinical practice: a review. *Gut*. 2001; 49 Suppl 4: iv2-10.
2. Carrera-Guermeur N, Martín-Crespo RM, Ramírez HJ, Pantoja Á, Luque-Mialdea R. Octreotide and enterocutaneous fistula closure in neonates and children. *Eur J Pediatr*. 2016; 175(3): 305-12.

3. Kwon SH, Oh JH, Kim HJ, Park SJ, Park HC. Interventional management of gastrointestinal fistulas. *Korean J Radiol.* 2008; 9(6): 541-9.
4. Lisle DA, Hunter JC, Pollard CW, Borrowdale RC. Percutaneous gelfoam embolization of chronic enterocutaneous fistulas: report of three cases. *Dis Colon Rectum.* 2007; 50(2): 251-6.
5. Terzi MC, Agalar C, Habip S, Canda AE, Arslan NC, Obuz F. Closing Perianal Fistulas Using a Laser: Long-Term Results in 103 Patients. *Dis Colon Rectum.* 2018; 61(5): 599-603.
6. Mauri G, Sconfienza LM, Fiore B, Brambilla G, Pedicini V, Poretti D, et al. Post-surgical enteric fistula treatment with image-guided percutaneous injection of cyanoacrylic glue. *Clin Radiol.* 2013; 68(1): 59-63.
7. Jindal T, Sharma RK, Kamal MR, Mandal SN, Mukherjee S, Karmakar D. Successful management of urethro-ileal pouch fistula by cyanoacrylate glue instillation. *J Pediatr Urol.* 2012; 8(1): e13-5.
8. Tekkis PP, Fazio VW, Remzi F, Heriot AG, Manilich E, Strong SA. Risk factors associated with ileal pouch-related fistula following restorative proctocolectomy. *Br J Surg.* 2005; 92(10): 1270-6.