



LETTERS TO THE EDITOR

Cystic duct remnant syndrome as a cause of post-cholecystectomy syndrome[☆]



Síndrome del muñón cístico como causa de síndrome poscolecistectomía

To the Editor:

We present the case of a 59-year-old woman, with no history of interest, except for surgery for biliary colic 7 years previously in another hospital, where she underwent laparoscopic cholecystectomy converted to open surgery. She was seen for recurrent episodes of colicky pain in the last few months, occasionally related with eating, which began in the back and radiated to the right epigastrium and hypochondrium, with an accompanying vagal reaction. Initially suspecting a biliary rather than gastroduodenal origin, blood tests with liver profile, ultrasound and gastroscopy were performed, all of which were normal. Due to the persistence of symptoms, biliary magnetic resonance cholangiography (MRC) was performed to rule out biliary anatomical abnormalities and residual choledocholithiasis, with normal findings. The study was completed with endoscopic ultrasound (EUS), observing a cystic duct stump of around 15 mm in length with a hyperechoic arciform structure with posterior acoustic shadowing measuring 4–5 mm in diameter in the proximal portion, with local inflammatory signs and cystic wall thickening (1.5 mm). In view of these findings and the suspicion of cystic duct remnant syndrome (CDRS), it was decided by consensus with the general surgery department to attempt calculus removal by endoscopic retrograde cholangiopancreatography (ERCP). In this procedure, a Fogarty balloon probe is passed through the endoscope to pressure-inject contrast medium into the cystic duct remnant. Calculi were not initially observed in the stump, but after filling the entire structure with contrast medium following guide wire cannulation, the presence of an irregular filling defect at proximal level of at least 5–7 mm in diameter was confirmed; however, endoscopic

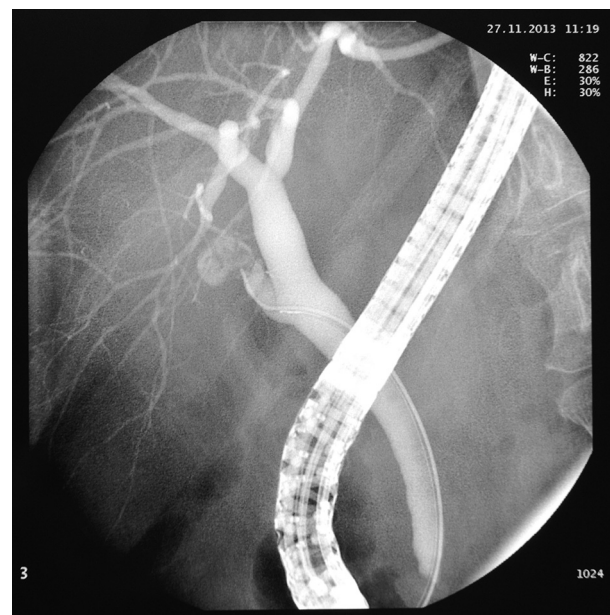


Figure 1 Cannulation of the cystic duct.

extraction was not possible (Figs. 1 and 2). Open surgery was then performed, finding a cystic duct stump embedded in a fibrotic mass in the gallbladder resection bed; the stump was mobilized and opened, revealing calculi within, and the cystic remnant was resected (Fig. 3). The patient progressed satisfactorily and currently remains asymptomatic.

It is currently estimated that between 5% and 40% of patients who undergo cholecystectomy can present episodes of abdominal pain. These can be similar to the symptoms that led to this indication, or appear after the intervention, and are grouped under the term “post-cholecystectomy syndrome” (PCS).¹ As this syndrome tends to become chronic and therefore extremely costly in terms of healthcare resources, a good diagnostic approach is crucial.

The aetiology of this syndrome is very diverse, and it can be secondary to biliary causes (organic and functional), such as residual choledocholithiasis, bile duct stenosis or leakage, neuroma in the scar tissue, biliary dyskinesia and Oddi sphincter dysfunction, among others. It is also essential to assess extra-biliary diseases not suspected before the surgery.² CDRS is one of the least common organic biliary disorders, with a prevalence of less than 2.5% in patients who have undergone cholecystectomy.

The cystic duct remnant is defined as a residual cystic duct greater than 1 cm in length, which in the presence of

[☆] Please cite this article as: Ruiz-Clavijo García D, Vila Costas J, Prieto Martínez C, Elizalde Apesteguía I, Tarifa Castilla A, Mercado Gutiérrez M, et al. Síndrome del muñón cístico como causa de síndrome poscolecistectomía. Gastroenterol Hepatol. 2016;39:722–724.

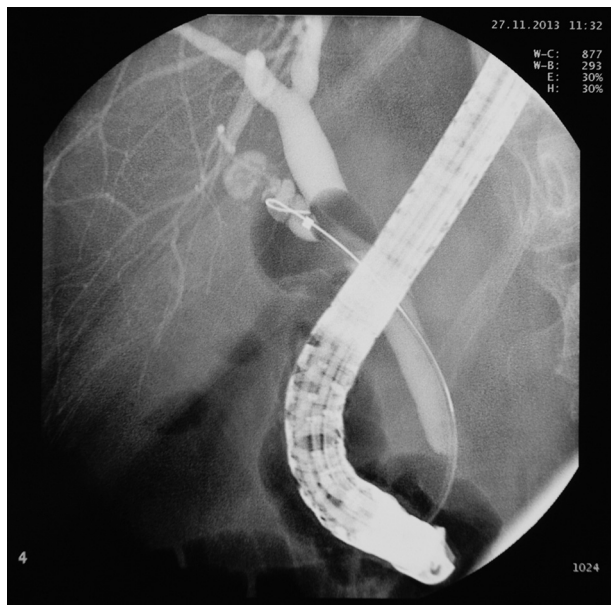


Figure 2 Cholangiography with Fogarty balloon and calculi in the remnant.

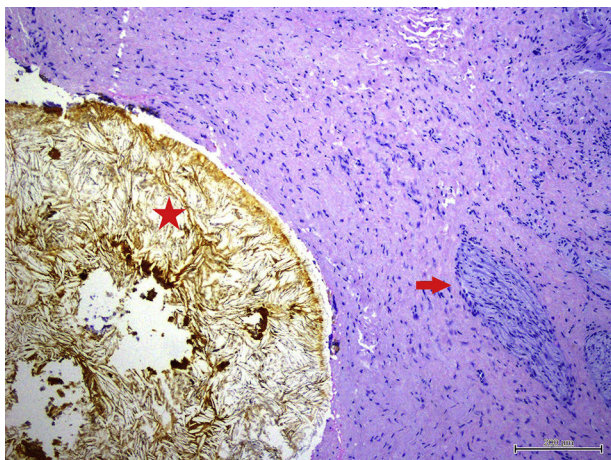


Figure 3 Histological image of the cystic duct with calculi (asterisk), fibrosis and hyperplasia of nerve plexuses (arrow).

calculi may produce symptoms.³ CDRS can present in the immediate postoperative period or even years after surgery. Its clinical presentation can mimic biliary colic produced by distension of the cystic remnant, sometimes accompanied by abnormal liver function tests and/or biliary obstruction.

It is important to note that the increasingly widespread use of laparoscopic gallbladder surgery has led to an increase in iatrogenic bile duct injuries. Most bile duct injuries are due to poor interpretation of the anatomy, as well as the presence of anatomical variants. The prevention of CDRS should be based on proper dissection and anatomical identification of the junction of the cystic and bile ducts.

The main diagnostic element in PCS is a good anamnesis and medical history, investigating previous symptoms and indication for the cholecystectomy, as well as details related with the procedure. Extra-biliary causes including irritable bowel syndrome, gastro-oesophageal reflux, peptic ulcer and chronic pancreatitis must first be ruled

out, as these are the most common causes of PCS. The aforementioned organic and functional biliary abnormalities should then be assessed. Full laboratory tests should be carried out, including liver profile, and when symptoms persist, complementary tests such as ultrasound and computed tomography should be done. When there is high suspicion of aetiology secondary to biliary abnormalities, MRC is the non-invasive technique of choice for evaluation of the biliary tree.⁴ An alternative to this is endoscopic ultrasound, which is basically more sensitive for the detection of choledocholithiasis <5 mm, with ERCP relegated to a strictly therapeutic technique.⁵

Once extra-biliary causes have been ruled out, and with compatible symptoms and imaging tests showing a long cystic remnant containing calculi, the diagnosis of CDRS should be made.

The therapeutic approach includes ERCP with sphincterotomy and possible stent placement to aid drainage of the main bile duct, together with definitive surgical treatment by resection of the bile duct remnant. This should be indicated in patients with recurrent symptoms and evidence of residual calculi in the remnant.^{6,7} The laparoscopic approach would be ideal, but the existence of fibrotic changes between the remnant and main bile duct can increase the risk of iatrogenic injury with respect to open surgery.⁸ Resection of the remnant seems to resolve symptoms in most cases. Another alternative to surgical and endoscopic treatment is extracorporeal shock wave lithotripsy, which has been successful in some published studies.⁹

Conflict of interests

The authors declare that they have no conflict of interests.

References

1. Perea E, Bhatt S, Dogra VS. Cystic duct remnant syndrome. *J Clin Imaging Sci.* 2011;1:2.
2. Jaunoo SS, Mohandas S, Almond LM. Postcholecystectomy syndrome. *Int J Surg.* 2010;8:15–7.
3. Shaw C, O'Hanlon DM, Fenlon HM, McEntee GP. Cystic duct remnant and the "post-cholecystectomy syndrome". *Hepatogastroenterology.* 2004;51:36–8.
4. Girometti R, Brondani G, Cereser L, Como G, del Pin M, Bazzocchi M, et al. Post-cholecystectomy syndrome: spectrum of biliary findings at magnetic resonance cholangiopancreatography. *Br J Radiol.* 2010;83:351–612.
5. Mohamadnejad M, Hashemi SJ, Zamani F, Baghai-Wadji M, Malekzadeh R, Eloubeidi MA. Utility of endoscopic ultrasound to diagnose remnant stones in symptomatic patients after cholecystectomy. *Endoscopy.* 2014;46:650–5.
6. Márquez Rojas J, Medina Ortiz JA, Martínez Mateo YA, Matito Díaz MJ, Arribas Jurado M. Cystic duct remnant syndrome: myth or reality. *Cir Esp.* 2014;92:496–7.
7. Pradeep C, Anil S, Amit G, Yusuf A, Khoobsurat N, Manish B, et al. Residual gallbladder stones after cholecystectomy: a literature review. *J Minim Access Surg.* 2015;11:223–30.
8. Parmeggiani D, Cimmino G, Cerbone D, Avenia N, Ruggero R, Gubitosi A, et al. Biliary tract injuries during laparoscopic cholecystectomy: three case reports and literature review. *G Chir.* 2010;31:16–9.

9. Judd S, Miller L, Antaki F. Symptomatic calculi in a remnant gallbladder: a rare cause of post-cholecystectomy syndrome and biliary pancreatitis. *Endoscopy*. 2014;46 Suppl. 1:E67–8.

David Ruiz-Clavijo García^{a,*}, Juan Vila Costas^a, Carlos Prieto Martínez^a, Inmaculada Elizalde Apesteguía^a, Antonio Tarifa Castilla^b, María Mercado Gutiérrez^c, Marian Casi Villarroja^a, Jesús María Urman Fernández^a, Francisco Javier Herrera Cabezón^b

^a Servicio de Aparato Digestivo, Complejo Hospitalario de Navarra, Pamplona, Navarra, Spain

^b Servicio de Cirugía General, Complejo Hospitalario de Navarra, Pamplona, Navarra, Spain
^c Servicio de Anatomía Patológica, Complejo Hospitalario de Navarra, Pamplona, Navarra, Spain

* Corresponding author.

E-mail address: davidruizcla@gmail.com

(D. Ruiz-Clavijo García).

2444-3824/

© 2016 Elsevier España, S.L.U., AEEH and AEG. All rights reserved.

Foot drop during adalimumab therapy for Crohn disease[☆]



Desarrollo de pie caído durante el tratamiento con adalimumab en enfermedad de Crohn

To the Editor:

Common peroneal nerve (CPN) palsy is the most common lower extremity mononeuropathy, but it is a rare complication of anti-tumour necrosis factor- α (anti-TNF- α) drugs.

We present the case of a 44-year-old man who had been diagnosed with Crohn disease, with extensive small intestine involvement (Montreal classification A2, L1+L4, B1), more than 8 years previously. At the time of consultation he was in remission, and had been taking adalimumab 40 mg every 14 days as monotherapy for the last 4 years. After multiple sclerosis (MS) had been ruled out by neurology specialists using the McDonald criteria, he attended the rehabilitation clinic for left foot drop (loss of dorsiflexion and eversion of the ankle) that had commenced 3 months earlier, with no history of trauma. Directed physical examination revealed a motor deficit in the dorsal flexion and eversion of the left ankle (1/5 on the modified Medical Research Council scale for muscle strength), and he received an ankle-foot orthosis for ambulation. An electromyogram showed severe neurapraxia-axonotmesis of the left CPN with no voluntary activity in the CPN-dependent muscles. In view of these findings, rehabilitation treatment was started, which consisted of 21 sessions of electrostimulation and 9 of kinesitherapy. An improvement in muscle balance was noted in the re-evaluation after this intervention: muscle balance was 3/5, and the patient was able to walk independently with no technical aids, so it was not necessary to modify the biological treatment of his underlying disease.

The question of whether neurological complications are part of the spectrum of extra-intestinal manifestations of inflammatory bowel disease, or simply secondary to nutritional deficiencies or medication use, is widely debated nowadays.¹ These complications are rarely associated with

anti-TNF- α drugs—incidence is estimated to be 0.1% for adalimumab and 0.03% for infliximab.² Of these complications, the most commonly described are development or exacerbation of MS and demyelination of the central nervous system (CNS), with peripheral nerve demyelination being even more uncommon. Furthermore, a clear cause-effect relationship cannot always be established between neuropathies and the use of anti-TNF agents. In a study to evaluate the onset of complications over time in a series of 9 patients who received anti-TNF- α (4 for rheumatologic disorders, 3 for sarcoidosis, 1 for psoriasis and 1 for Crohn disease), most developed neurological deficits within the first 6 months of treatment, although in the case of the Crohn disease, these appeared 38 months after initiation of infliximab.³

Certain predisposing factors may contribute to the onset of CPN palsy, such as weight loss, trauma or continuous compression (habit of crossing one's legs).⁴ Electromyography is the gold standard for diagnosis.

Although it has been established that the use of anti-TNF- α should be avoided in patients with a history of MS or demyelinating diseases of the CNS,^{5,6} there is no consensus on their use when neurological disorders develop during administration of these drugs. The course of the neuropathy is usually unpredictable, so the risk of continuing anti-TNF treatment should be weighed up on an individual basis against the benefit it provides to the underlying disease.⁸ Both maintenance of treatment³ and a reduction in the anti-TNF dose have been described, so discontinuation of this drug should be limited to patients with controlled underlying disease and incapacitating neuropathy.⁷

Other therapeutic options in the case of peripheral demyelinating neuropathies are corticosteroids, cyclophosphamide, intravenous immunoglobulins or plasmapheresis. In the case presented, the improvement in the Crohn disease under immunomodulatory treatment prompted us to choose conservative management. This achieved a good outcome, and therefore could be a therapeutic option in similar individual cases.

References

1. Gondim Fde A, de Oliveira GR, Teles BC, Aquino Pde S, Brasil ÉF, Carvalho AM, et al. Clinical and electrodiagnostic findings in patients with peripheral neuropathy and inflammatory bowel disease. *Inflamm Bowel Dis*. 2015;21:2123–9.

[☆] Please cite this article as: Collazo Diéguez M, González Carrera V. Desarrollo de pie caído durante el tratamiento con adalimumab en enfermedad de Crohn. *Gastroenterol Hepatol*. 2016;39:724–725.