

Case Report

Surgical Treatment of Lemmel's Syndrome - Contribution of Three New Cases Operated and Review of the Literature

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Abstract

Duodenum is the most common location of small bowel diverticulum. Most duodenal diverticula are solitary, acquired, asymptomatic and do not require treatment. Only 5% have some complications as intermittent obstructive jaundice in the absence of choledocholithiasis (Lemmel's syndrome). Endoscopic sphincterotomy is the most common treatment, especially in patients at high surgical risk. When endoscopic sphincterotomy fails, the most appropriate surgical treatment remains controversial.

Three new cases of Lemmel's syndrome operated and other 37 cases treated surgically are revised. Diverticulectomy is the most performed surgical technique, although is advisable to associate a sphincteroplasty in patients without previous endoscopic sphincterotomy to avoid recurrence of symptoms. As an alternative to diverticulectomy, different digestive and/or bile derivations have been performed with good results and individualized indications. Minimally invasive surgery is a valid approach in patients with Lemmel's syndrome, but until now limited to diverticula in the lateral wall of the duodenum.

Keywords: Lemmel's syndrome, Duodenal diverticula; Surgery; Endoscopic sphincterotomy

Introduction

Duodenum is the most common location of small bowel diverticulum [1]. However, it is difficult to determine the exact incidence of duodenal diverticula (0.16% to 22%) [2-6]. Most are solitary and located in the second duodenal portion [2,7]. Duodenal diverticula are usually asymptomatic, accidentally diagnosed and do not require treatment [2,7-9]. Approximately 10% refer abdominal pain and only 5% have some complications [1,8,10,11]. In 1934 Lemmel's Syndrome (LS) was described as intermittent obstructive jaundice in the absence of choledocholithiasis due to a duodenal diverticulum [12]. Since then, more than 200 cases of LS have been reported in the literature. The treatment is controversial, with numerous surgical techniques employed. We present three new cases of operated extraluminal duodenal diverticula and conduct a review of the surgical treatment described in the literature.

Clinical Cases

From March 2012 to February 2020, we have treated four patients with LS. The first case is a 69-year-old male with a history of valvulopathy with aortic valve prosthesis and ventricular dysfunction, in treatment with warfarin. The patient presents episodes of recurrent cholangitis. Computed Tomographic (CT) scan shows cholelithiasis without dilation of the bile duct and a 4 cm diverticulum in second juxtapancreatic duodenal portion. Endoscopic Retrograde

Cholangiopancreatography (ERCP) and subsequent surgery are proposed but the patient rejects any interventionism. After two years of follow-up, the patient is asymptomatic.

The other three patients were operated on and are described below.

Case 1

A 69-year-old woman who presents epigastric pain with occasional vomiting. The laboratory tests are normal and the ultrasound shows a gallbladder with stones inside and dilation of the intra and extrahepatic bile duct. A Magnetic Resonance Cholangiopancreatography (MRCP) is performed confirming the presence of cholelithiasis, with a maximum Common Bile Duct (CBD) of 14 mm and a 38 mm diverticulum in the medial aspect of the second duodenal portion. Duodenoscopy shows a large duodenal diverticulum with solid food content and an intradiverticular papilla, which cannot be properly cannulated. Endoscopic ultrasound is performed appreciating a 12 mm CBD without choledocholithiasis. The patient is operated through a right subcostal incision. Cholelithiasis and dilation of the CBD are seen. Cholecystectomy and duodenostomy are performed, obtaining detritus inside a diverticulum in the second duodenal part. The intervention is completed with an antrectomy, gastrojejunostomy and a Roux-en-Y choledochojejunostomy. On the 8th postoperative day the patient is discharged without complications and after five years of follow-up, is asymptomatic.

Case 2

A 73-year-old woman with a pacemaker and a history of triple valve replacement (mitral, aortic and tricuspid), in treatment with warfarin. Presents fever and abdominal pain in the right upper quadrant. In laboratory tests there are 11,000 leukocytes, and total bilirubin of 2.5 mg/dL. Ultrasound shows a gallbladder without cholelithiasis and an 8 mm CBD. A CT scan is performed where a distended gallbladder is seen without signs of cholecystitis or stones and a 20 mm juxtapancreatic duodenal diverticulum. The duodenoscopy confirms the presence of a 2 cm diverticulum in the second duodenal portion and a normal-looking patent papilla so that an Endoscopic Sphincterotomy (ES) is

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not performed. Endoscopic ultrasound discards choledocholithiasis and appreciates slight compression of distal CBD by a duodenal diverticulum. The patient is surgically operated on. After a Kocher maneuver, a duodenal diverticulum is seen. Cholecystectomy is performed with the introduction of a transcystic Fogarty catheter appreciating the papilla in the margin of the duodenal diverticulum, so it is decided to perform a side-to-side choledochoduodenostomy. The patient is discharged on the 6th postoperative day and after two months of follow-up is asymptomatic.

Case 3

A 73-year-old woman who presents episodes of colic pain in the right upper quadrant. There are no alterations in the laboratory tests and the ultrasound shows a distended gallbladder with cholelithiasis and a 9 mm CBD without evidence of an obstructive cause. MRCP shows a sharpening of the intrapancreatic portion of the distal CBD, without lithiasis inside, suggestive of benign stenosis. CT confirms the presence of a juxtapapillary duodenal diverticulum as a possible cause of CBD dilation. In the barium study, a duodenal diverticulum is evident on the medial slope of the second duodenal portion. An ERCP is attempted, being impossible to cannulate the papilla during the duodenoscopy. The patient is operated on performing a cholecystectomy and diverticulectomy. Due to the recurrence of the symptoms, surgical reintervention is decided one year after the first surgery. Transduodenal sphincteroplasty and section of the bile duct and duodenum are performed along with a duodenojejunostomy and a Roux-en-Y choledochojejunostomy. The patient is discharged on the 20th postoperative day after infection of the surgical site. After 13 months the patient is asymptomatic.

Discussion

Duodenum is the most common location of small bowel diverticulum [1]. Most are solitary and 60% are present in the second duodenal portion (88% are medial, 4% lateral and 8% posterior) [2,7-9,13]. Almost 90% are acquired and extraluminal by a herniation of the mucosa and submucosa through a defect in the muscle wall of the duodenum [7]. Intraluminal diverticula are protrusions of the mucosa and submucosal towards the duodenal lumen, of congenital origin [7]. The incidence of duodenal diverticula is variable depending on the different diagnostic methods used: 5.2% to 22% in autopsies, 2% to 5% in upper GI barium studies and 7% to 20% in ERCP [1,2-6,14]. Depending on the relationship with the papilla, duodenal diverticula are divided into three types: type I with the intradiverticular papilla; type II (juxtapapillary) with papilla in the margin of the diverticulum and type III (periampullary) in which the papilla is near the diverticulum (less than 3 cm) [2,15]. They are most common in women and after the sixth decade [4,7,15-17]. Most are asymptomatic, accidentally diagnosed and do not require treatment. Approximately 10% have epigastric or right upper quadrant pain, mainly postprandial and only 5% have some complications: diverticulitis, bleeding, perforation or fistula and pancreaticobiliary complications such as cholelithiasis, choledocholithiasis, cholangitis, acute pancreatitis and obstructive jaundice [1,7,10,11].

In 1934 Lemmel's syndrome was described as intermittent obstructive jaundice in the absence of choledocholithiasis due to a duodenal diverticulum, usually from the second duodenal portion, although there is some described case produced by a giant diverticulum in the third portion of the duodenum [12,18]. Most are extraluminal diverticula, being intraluminal very exceptional [7,19,20]. Miyazaki et al. [21] demonstrated that duodenal diverticula

can be mechanically distended and compressed the distal CBD [21]. Other pathogenic factors described are the presence of an enterolith or bezoar inside the diverticulum, diverticulitis, fibrosis of papilla secondary to chronic mechanical irritation or motility dysfunction of the sphincter of Oddi with decreased basal pressure [11,22,23]. The incidence of LS is higher in patients with a juxtapapillary diverticulum [14,24,25]. The size of the diverticulum and the average age of SL patients are larger than those with duodenal diverticula without LS [14]. Clinically LS is characterized by jaundice, mild colic abdominal pain or acute cholangitis [25]. Jaundice is not usually very striking, with an average elevation of total bilirubin of 4.2 mg/dL [14]. The first diagnostic imaging technique is an ultrasound that usually shows dilation of the bile duct without an obvious cause. CT scan and MRCP allow to rule out the presence of CBD stones and to demonstrate the diverticular neck, as well as its relationship with papilla [26].

Only symptomatic duodenal diverticula require treatment. The most common indications of surgery are bleeding and perforation of the diverticulum [16]. In patients with LS the management is controversial and depends mainly on an individualized assessment of each case. When bile duct obstruction is caused by duodenal diverticulitis, favourable results have been achieved with antibiotic treatment [22]. Duodenoscopy allows food debris or bezoar to be removed inside the diverticulum, confirm the size of the diverticulum, the relationship with the papilla and perform an endoscopic ultrasound to rule out possible small CBD stones not previously diagnosed [7,11]. In intraluminal diverticula, surgical or endoscopic excision may be performed [19,27,28], although in our review there have been no intraluminal diverticula. Historically until development and increased experience gained with ERCP, surgical treatment has been the most common (62.5%) [18]. However, endoscopic treatment of LS has increased in recent decades. Endoscopic Sphincterotomy (ES) may be the definitive treatment when there is purulent cholangitis, fibrosis of papilla and in patients at high surgical risk, especially in type I and II duodenal diverticula [14,24,25,29,30]. In a review of 1923 ERCP in patients with cholangitis, 17.5% had duodenal diverticula and LS was diagnosed in 11.6% of cases [14]. Cannulation difficulties were greatest in patients with an intradiverticular papilla and 15.4% of cases a bezoar impacted in a duodenal diverticulum were found. When ES fails or the size of the diverticulum is large, surgical treatment is indicated [7,31]. We have analyzed a total of 40 patients described in the literature surgically treated as a result of LS (Table 1), after excluding those cases in which the technique performed was not specified. Women are predominant (71.4%) and the average age is 70.3 years (54-81). In seven cases (17.5%) diverticuloplasty has been performed but the complication rate has been high [27,32].

Diverticulectomy is the most performed surgical technique in the treatment of duodenal diverticula since Cattell and Mudge published in 1952 the first 24 cases performed in the same centre [33,34]. In 1972 Pinotti et al. [35] reported very good results in 12 patients with diverticula treated with diverticulectomy (11 associated sphincteroplasties). However, relapse of symptoms is common, and the mortality of diverticulectomy described by other authors is high (3% to 13%), not only in cases operated by urgent complications but also in elective surgeries [2,7,8,10,31,34,36,37]. In bleeding and perforations of diverticula located in the lateral or anterior wall of the duodenum diverticulectomy is a valid option [17], although in LS diverticula in the medial wall of the duodenum are more common and the surgical technique is different. After a Kocher maneuver, the location of the papilla and its relationship to the diverticulum is determined, usually

Table 1: Surgical treatment of Lemmel’s syndrome.

Author (year)	Cases	Age	Sex	Symptoms	Previous ES	Surgical technique	Mortality	Complications	Follow-up	Recurrence	Reop.
Neill (1965)	1	63	M	Jaundice	No	Diverticulectomy + T-tube	No	No	NS	No	No
Willox (1969)	3	NS	NS	NE	No	Diverticuloplasty + T-tube	NS	NS	NS	NS	NS
	2	NS	NS			ChD	NS	NS	NS	NS	NS
McSherry (1970)	1	69	F	Cholangitis	No	Diverticulectomy + T-tube	No	No	9 months	No	No
Sykes (1970)	1	76	M	Jaundice	No	Roux-en-Y Cholecistojunostomy	No	No	1 year	No	No
Pinotti (1971)	1	NS	NS	Cholangitis	No	Diverticulectomy	No	No	10 years	Yes	No
	1	NS	NS	Cholangitis	No	Diverticulectomy + T-tube + sphincteroplasty	No	No	4 years	No	No
Solhaug (1974)	1	58	F	Cholangitis	No	Sphincteroplasty	No	No	NS	No	No
Donald (1979)	1	66	F	Pain	No	ChD	No	No	7 years	No	No
Alfonsi (1979)	1	72	F	Jaundice	No	Diverticulectomy + T-tube	No	No	NS	No	No
Critchlow (1985)	1	80	F	Cholangitis	No	Roux -en-Y DJ + T-tube	No	No	18 months	No	No
Gudjonsson (1988)	1	56	F	Pain	No	Roux-en-Y CJ	No	No	NS	No	No
Trondsen (1990)	4	NS	NS	Cholangitis	No	Diverticuloplasty + sphincteroplasty	No	Diverticular bleeding Retroperitoneal hematoma CBD perforation	3-10 years	No	No
Van der Linde (1997)	1	72	F	Pain + Jaundice	No	Roux-en-Y DJ	No	No	25 months	No	No
	1	58	F	Pain + Jaundice	No	Diverticulectomy	No	No	30 months	No	No
Vassilakis (1997)	1	NS	3M / 2F	Cholangitis	NS	Roux-en-Y DJ	No	Anastomotic ulcer	NS	No	Antrectomy + Roux-en-Y GJ + TVG
	4	NS		Cholangitis	NS	Roux-en-Y DJ + CJ	No	No	1 year	No	No
Castilho (2003)	1	74	F	Cholangitis	No	Roux-en-Y CJ + T-tube	No	No	2 years	No	No
Yoneyama (2004)	1	72	M	Pain + Jaundice	No	Diverticulectomy	No	Choledocholithiasis	33 months	No	ES
	1	77	F	Cholangitis	No	Diverticulectomy + T-tube	No	No	31 months	No	No
	1	81	F	Pain + Jaundice	No	Diverticulectomy + Transcystic catheter	No	No	22 months	No	No
Mathis (2007)	1	NS	NS	Jaundice	NS	Diverticulectomy	No	Duodenal stenosis	NS	NS	No
Kellas (2010)	1	78	F	Pain + Jaundice	No	Diverticulectomy + robotic ChD	No	Diarrhea (<i>clostridium</i>)	18 months	No	No
Perrot (2012)	1	81	F	Cholangitis	Yes	Biliodigestive derivation	No	No	NS	No	No
Karayannakis (2012)	1	72	F	Jaundice	No	Diverticulectomy	No	No	1 week	No	No
Beisani (2013)	1	75	F	Pain	Yes	Roux-en-Y CJ	No	No	NS	No	No
Carmona (2017)	1	54	M	Cholangitis	No	Diverticulectomy + Roux-en-Y CJ	No	Biliary leak + Subphrenic abscess	2 years	No	No
Khan (2017)	1	69	M	Pain + Jaundice	No	Diverticulectomy	No	NS	NS	No	No
Castellón (2020)	1	69	F	Pain	No	Antrectomy + GJ + Roux-en-Y CJ	No	No	5 years	No	No
	1	73	F	Cholangitis	No	ChD	No	No	2 months	No	No
	1	73	F	Pain	No	Diverticulectomy	No	Surgical site infection	13 months	Yes	Sphincteroplasty + DJ + Roux-en-Y CJ

M: Male; F: Female; NS: Not Specified; ES: Endoscopic Sphincterotomy; ChD: Choledocoduodenostomy; DJ: Duodenojejunostomy; CJ: Choledochojejunostomy; Reop: Reoperation; GJ: Gastrojejunostomy; CBD: Common Bile Duct; TVG: Truncular Vagotomy

by an anterior duodenotomy or using a Fogarty catheter inserted through the bile duct into the duodenum [37]. The diverticulum is identified, everted with a Babcock clamp, dissected, avoiding injury to the pancreas and the defect is sutured [35]. Diverticulectomy has been performed alone or with another associated technique, in 17 patients (42.5%) with LS. The mortality of diverticulectomy in these cases

has been zero and the complications minimal. However, in 11.7% of cases the symptoms relapsed, probably because diverticulectomy does not derive food content at the diverticulum level and because none of these has been given a previous ES. Therefore, we consider that when performing a diverticulectomy it is advisable to associate a sphincteroplasty if an ES has not been previously performed.

Diverticulectomy in SL is not without the risk of injury to the Wirsung duct or distal CBD, and can sometimes be very complicated to perform, especially in large diverticula embedded deep within the head of the pancreas and highly vascularized by an associated chronic inflammatory process [27]. In these cases, different digestive and/or bile derivations have been proposed as an alternative to diverticulectomy. Until the late 1970s, more resections and diverticuloplasties were performed. Afterwards, with the development of the ES, more surgical bypass techniques have been performed. Derivations should be avoided in patients with recurrent pancreatitis or stenosis of the ampulla [31,38]. In 19 patients with LS (47.5%) some type of derivation has been performed (two cases with a diverticulectomy associated). Five side-to-side choledochoduodenostomies (12.5%) have been performed [17,32,39]. It is a simpler, faster and less aggressive technique, so it would be more suitable in older patients and associated comorbidity, with poorly dilated bile duct and in patients with previous gastrectomies [7,40]. Manny et al. [38] have performed an antrectomy, vagotomy and Billroth II gastrojejunostomy after the failure of a choledochoduodenostomy in two patients with duodenal diverticula, although this option leads to potential long-term functional complications [31]. Choledochojejunostomy is preferable when the CBD is dilated and has been performed with good results in five other patients (12.5%) [9,10,40,41]. Critchlow et al. [31] propose to perform an end-to-side Roux-en-Y duodenojejunostomy as a treatment for complicated duodenal diverticula. In LS this technique has been performed in three cases, developing one patient an anastomotic ulcer that required surgical reintervention [31,42,43]. To reduce the risk of ulceration and derive bile flow, Vassilakis et al. [43] have associated a choledochojejunostomy to a Roux-en-Y duodenojejunostomy in four other patients with LS (10%) with good results. There are 15 patients with duodenal diverticula treated by minimally invasive surgery with low morbidity, although only one (7%) was operated for LS due to a diverticulum in the lateral wall of the duodenum.

Conclusion

In conclusion, Lemmel's syndrome is a rare complication of duodenal diverticula and treatment is challenging. The most common treatment is endoscopic sphincterotomy. When it fails, diverticulectomy and biliodigestive derivations, with individualized indications, are the most commonly used surgical techniques.

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