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Systematic Review

Biliary Leak from Ducts of Luschka: Systematic Review of the Literature

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Abstract: Injury to the Luschka ducts (LDs), also named "subvesicular" ducts, is an under-reported cause of biliary leak following laparoscopic cholecystectomy (LC). A systematic literature search according to PRISMA guidelines was conducted in PubMed, EMBASE and Cochrane Library including all publications that described a bile leak from an LD. A total of 136 articles were retrieved from the searched databases. After the removal of duplicates and non-eligible papers, 48 studies reporting 231 leaks were included: 20 (41.6%) case reports, 2 (4.3%) comparative studies, 7 (14.9%) meeting abstracts and 19 (40.4%) retrospective cohort articles. The rate of LD leak ranges from 0.05% to 1.9%, but injury to a duct of Luschka was the second most common cause of biliary leakage in all the cohort studies (5.5% to 41%). In 21 (43.7%) cases, the leak was successfully treated with a sphincterotomy through Endoscopic Retrograde Cholangiopancreatography (ERCP) plus or minus stenting, and in 12 (25%), re-laparoscopy was necessary.

Keywords: Luschka duct; biliary tree injury; biliary leak; cholecystectomy

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1. Introduction

Laparoscopic cholecystectomy (LC) is one of the most performed procedures in general surgery worldwide [1]. Bile duct injury (BDI) is the most feared complication of LC, and its incidence is reported to be 0.3–0.7% [2]. Several classifications for BDI have been proposed [3]; one of the most recognized is Strasberg's classification of BDI, which mainly addresses bile leaks which are more commonly seen in the laparoscopic era. Bile leaks may arise from a variety of sites, mainly from cystic stumps, Luschka ducts (LDs) or other main bile ducts [4].

The leakage of Luschka ducts, also named "subvesicular" ducts, along with cystic stump leaks (together classified as Strasberg A) are the most common locations of BDI after LC [5]. LDs were first described by Hubert von Luschka, a German anatomist, in 1863, and in more recent studies, its incidence varies from 1 to 50% due to different diagnostic methods [6]. LDs are usually confused with hepaticocholecystic ducts (HCDs), which are real variations of the biliary tree draining into the gallbladder [6]. LDs originate from the right hepatic lobe, along the gallbladder bed, and usually drain in the extrahepatic bile ducts. Some authors have also proposed a common classification for LDs and HCDs: (1) segmental or sectorial subvesical bile ducts, (2) accessory subvesical bile ducts, (3) hepaticocholecystic bile ducts, and (4) aberrant subvesical bile ducts [7].

Despite the classification methods on these aberrant ducts, the literature mainly relies on case reports [8,9]. To the best of our knowledge, only one case of intraoperative diagnosis has been associated with proper video imaging [10].

The primary aim of our study was to review all reported cases of leaks from LDs.

2. Materials and Methods

2.1. Search Strategy

The present review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [11]. PubMed, EMBASE and Cochrane Library databases were searched using the following terms: "duct of Luschka" AND "leak". No temporal interval was set. References were further reviewed to find other articles of interest.

2.2. Eligibility Criteria

All types of studies (case reports, case series, comparative, retrospective, prospective, meeting abstracts) in the English language or translated to English reporting cases of bile leakage from an LD were included in our review. Duplicates, previous reviews, and papers on biliary tree injuries not reporting cases of leakage from LDs were excluded.

2.3. Data Extraction

Two authors independently extracted the following data from the extracted articles: first author name, year of publication, type of study, total number and rate of LD leaks, rate of LD leaks/overall cases of leaks, type of intervention.

A dedicated table was filled in by the two authors with this information, and a third author compared and reviewed their results.

3. Results

A total of 136 articles (45 in PubMed, 90 in EMBASE and 1 in Cochrane Library) were retrieved from the searched databases. In total, 44 duplicates were removed, and, after title and abstract review, 43 studies were found to be eligible; another 5 studies were discovered in the references (Table 1) [6,8,9,12–57]. A PRISMA flowchart [58] is pictured in Figure 1.

Table 1. Summary of studies reporting leaks from Luschka ducts. PTC = transhepatic cholangiography; LC = laparoscopic cholecystectomy.

Authors	Type of Study	Leak (n/%)	DL/Other Causes of Leak (%)	Intervention	Treatment
Garcia et al. (2022) [6]	Case report	1	Not reported	LC	Re-laparoscopy
Crowley et al. (2021) [12]	Case report	1	Not reported	LC	Endoscopic
Sanchez et al. (2021) [13]	Poster presentation	1	Not reported	LC	Percutaneous drain
Nagaratnam et al. (2021) [14]	Poster presentation	2	7.4%	LC	Endoscopic
Hussain et al. (2020) [15]	Retrospective cohort study	4 (0.19%)	Not reported	LC	Re-laparoscopy
Chandra et al. (2020) [16]	Retrospective cohort study	23	41%	LC; Hepatic resection, liver trauma	Endoscopic

Table 1. *Cont.*

Authors	Type of Study	Leak (n/%)	DL/Other Causes of Leak (%)	Intervention	Treatment
Paramythiotis et al. (2019) [17]	Case report	1	Not reported	LC	Endoscopic
Tabanera et al. (2019) [9]	Case report	1	Not reported	Liver Transplant	Re-laparotomy
Masoni et al. (2018) [18]	Case report	1	Not reported	LC	Endoscopic
Kumar et al. (2018) [19]	Case report	1	Not reported	LC	Laparoscopic (intraoperative)
Lytvyn et al. (2019) [20]	Congress presentation	52	41.50%	LC	Percutaneous drain, endoscopic and re-laparoscopy
Antequera et al. (2019) [21]	Congress presentation	1	Not reported	LC	Re-laparoscopy
Abtar et al. (2018) [22]	Case report	1	Not reported	LC after bariatric surgery	Re-laparoscopy
Bonatti et al. (2017) [23]	Case report	1	Not reported	LC	Endoscopic
Ko et al. (2017) [24]	Case report	1	Not reported	LC	Endoscopic
Mariolis et al. (2017) [25]	Case report	1	Not reported	LC	Laparoscopic (intraoperative)
Aspinen et al. (2015) [26]	Comparative study	1 (1.9%)	Not reported	LC	Re-laparoscopy
Shahnawaz et al. (2015) [27]	Congress presentation	1	not reported	LC	Laparoscopic (intraoperative) + endoscopic
Salsano et al. (2015) [28]	Case report	1	Not reported	LC	Percutaneous transhepatic cholangiography (PTC)
Gutierrez et al. (2013) [29]	Case report	1	Not reported	LC after bariatric surgery	Endoscopic
Tewani et al. (2013) [30]	Retrospective cohort study	30	24%	LC	Endoscopic
Thayalasekaran et al. (2012) [31]	Congress presentation	11	13%	LC	Percutaneous drain, endoscopic and re-laparoscopy
Lo Nigro et al. (2012) [32]	Retrospective cohort study	2	5.5%	LC	Endoscopic
Tran et al. (2011) [33]	Congress presentation	1	Not reported	LC	Percutaneous + endoscopic
Hwang et al. (2011) [34]	Case report	1	Not reported	LC	Endoscopic
Fasoulas et al. (2011) [35]	Retrospective cohort study	4	6.06%	LC	Endoscopic
Neumann et al. (2010) [36]	Case report	1	Not reported	OC	Endoscopic
Pinkas et al. (2008) [37]	Comparative study	5	25%	LC	Endoscopic
Bledsoe et al. (2008) [38]	Case report	1	Not reported	LC	Endoscopic
Tantia et al. (2008) [39]	Retrospective cohort study	11	21%	LC	Endoscopic/Surgical

Table 1. *Cont.*

Authors	Type of Study	Leak (n/%)	DL/Other Causes of Leak (%)	Intervention	Treatment
Rulli et al. (2006) [40]	Case report	1	Not reported	LC	Re-laparoscopy
Ramia et al. (2005) [41]	Retrospective cohort study	2 (0.15%)	Not reported	LC	Re-laparoscopy
Misra et al. (2005) [42]	Retrospective cohort study	5 (0.5%)	45%	LC	Endoscopic, fibrin glue, re-laparoscopy
Kaffes et al. (2005) [43]	Retrospective cohort study	15	15%	LC	Endoscopic
Sandha et al. (2004) [44]	Retrospective cohort study	26	13%	LC	Endoscopic
Rossi et al. (2002) [45]	Retrospective cohort study	5 (2%)	Not reported	LC	Endoscopic
De Palma et al. (2002) [46]	Retrospective cohort study	4	6,3%	LC	Endoscopic
Albishri et al. (2001) [47]	Case report	1	Not reported	Liver Transplant	Re-laparotomy
Braghetto et al. (2000) [48]	Retrospective cohort study	3 (0.05%)	23%	LC	Re-laparotomy
Wills et al. (2000) [49]	Retrospective cohort study	10 (0.5%)	67%	LC	Re-laparoscopy
Jamshidi et al. (1999) [50]	Case report	1	Not reported	LC	Re-laparoscopy
Mergener et al. (1999) [51]	Retrospective cohort study	15	17%	LC; OC	Percutaneous drain, endoscopic, re-laparotomy and re-laparoscopy
Aru et al. [52]	Retrospective cohort study	1 (10%)	10%	LC	Endoscopic
Bour et al. (1997) [53]	Case report	1	Not reported	LC	Re-laparoscopy
Albasini et al. (1995) [54]	Retrospective cohort study	1 (0.2%)	Not reported	LC	Re-laparoscopy
Sefr et al. [55]	Retrospective cohort study	3 (0.2%)	37.5%	LC	Re-laparoscopy
Frakes et al. (1993) [56]	Case report	1	Not reported	LC	Endoscopic
Brooks et al. (1993) [57]	Retrospective cohort study	2 (0.3%)	22%	LC	Percutaneous drain, endoscopic, re-laparotomy and re-laparoscopy
Frakes et al. (1993) [56]	Case report	1	Not reported	LC	Endoscopic

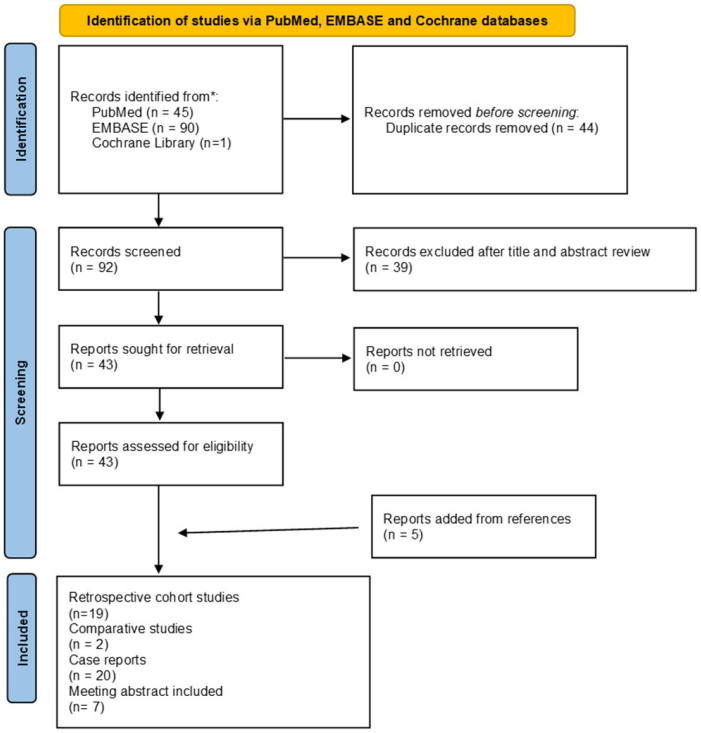

Figure 1. PRISMA flowchart of study selection.

3.1. Types of Studies

In total, 20 (41.6%) case reports, 2 (4.3%) comparative studies, 7 (14.9%) meeting abstracts and 19 (40.4%) retrospective cohort articles were retrieved in our search (Figure 2).

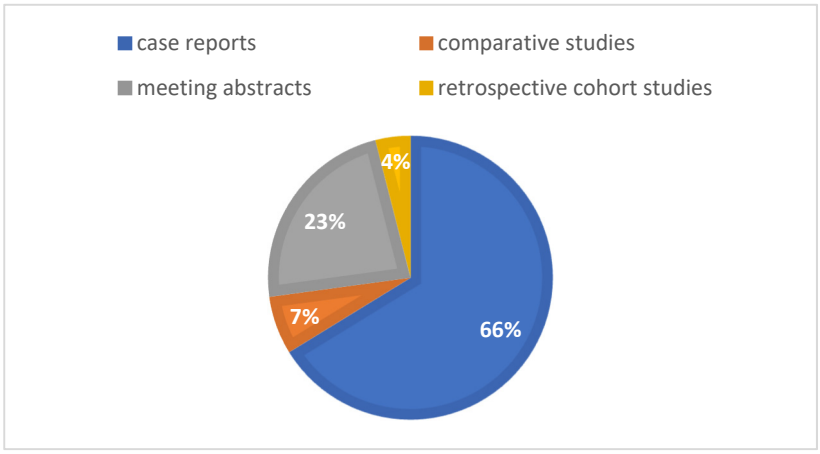

Figure 2. Pie chart of the included studies.

3.2. Rate of LD Leak

A total number of 231 leaks from ducts of Luschka were reported in the papers included in the present review. The rate of LD leak/intervention ranges from 0.05% to 1.9%, but injury to a duct of Luschka was the second most common cause of biliary leakage in all the cohort studies (5.5% to 41%) (Table 1).

3.3. Type of Intervention and Treatment

In 44 studies, the intervention was a cholecystectomy, which was laparoscopic (LC) in 41, open (OC) and LC in 1 and OC in 1. One study reported a leak after liver surgery, two after liver transplant and two after LC in patients with a previous history of bariatric surgery.

In 21 (43.7%) cases, the leak was successfully treated with a sphincterotomy through Endoscopic Retrograde Cholangiopancreatography (ERCP) plus or minus stenting, and in 12 (25%), re-laparoscopy was necessary; re-laparotomy was performed after open cases (3, 6.3%); in 2 (4.2%) patients, a percutaneous drain was resolutive; in 2 (4.2%), the leak was identified and treated intraoperatively; and in 8 (16.7%) studies, different types of treatment were reported (Figure 3).

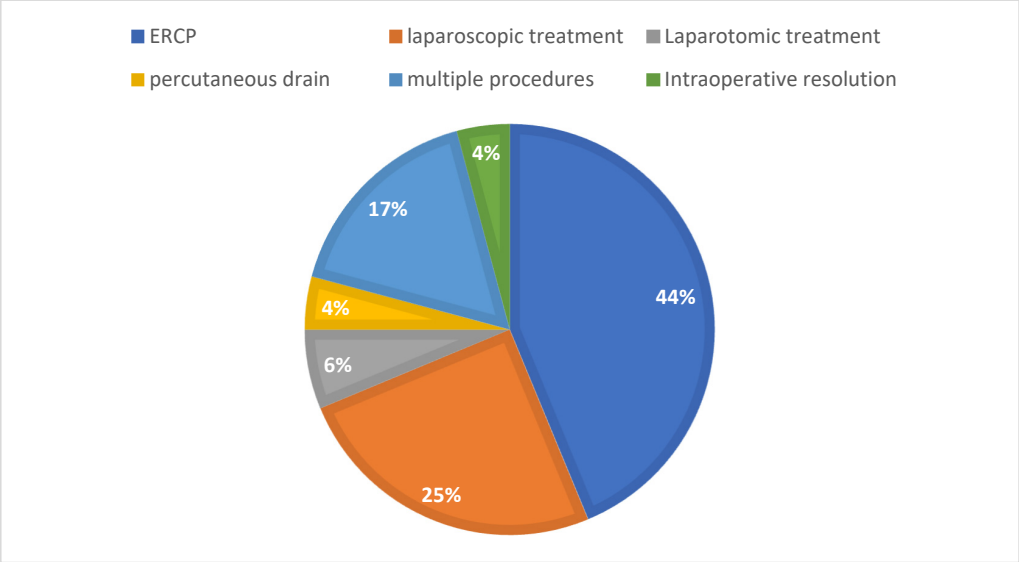

Figure 3. Pie chart of the treatments for LD leakage.

4. Discussion

4.1. Deliberation

BDI continues to be one of the most morbid complications following LC. During the open cholecystectomy era, it used to be 0.1–0.2%, but it increased to 0.4–0.6% in the laparoscopic era [59]. The diagnosis of BDI is most commonly diagnosed intraoperatively. Pekolj et al. [60] found that 90% of BDI cases are diagnosed intraoperatively. BDI is associated with increased morbidity and long-term decreased quality of life and loss of productivity [61]. It is of major importance to emphasize that LD leaks along with cystic stump leaks after LC have been shown to be associated with increased morbidity and mortality [62]. Therefore, if a BDI of any type is suspected, urgent diagnostic studies should be undertaken, and treatment should be started.

In the diagnostic imaging of BDI, an abdominal CT scan is recommended and magnetic resonance cholagiopancreatography (MRCP) can be used to localize the injury [63].

Our systematic review demonstrates that cases of intra- and postoperative leakage from a LDs are well documented in the modern literature. However, LD leaks are frequently presented as case reports.

Bile collection from subvesical duct injury is frequently diagnosed within the first postoperative week. Patients may report specific symptoms, such as abdominal or back pain, and signs such as abdominal wall tenderness and fever. An increase in serum bilirubin or transaminases is reported. This nonspecific symptomatology can be underestimated, and this could lead to serious consequences and to more serious complications (biliary peritonitis and sepsis). However, we also found cases in which symptoms arose weeks after cholecystectomy and cases where patients reported no symptoms at all. Generally, the severity of symptoms depends on the flow of bile into the abdomen and on the subsequent collection.

The management of LD leaks mainly depends on the presentation: those cases which are intraoperatively diagnosed are usually clipped during the procedure. However, despite the better view offered by the laparoscope, the intraoperative aspect of the liver bed

may appear normal, but a postoperative leak from an LD may occur. The postoperative manifestation of an LD leak is heterogenous and can be treated in several ways according to the clinical manifestation of the patient. In cases where the patient is septic, and a large amount of fluid is seen on imaging (Figure 4) with or without bile duct dilatation, we suggest surgical exploration for abdominal washout. It can be difficult to diagnose the exact location of an LD leak intraoperatively; therefore, we always recommend the assistance of an HPB surgeon and conversion to open surgery in difficult cases. In cases where a small biloma is seen on imaging and the patient is doing well with no signs of sepsis, conservative management can be taken with intravenous antibiotics, ERCP plus or minus stenting and follow-up imaging.

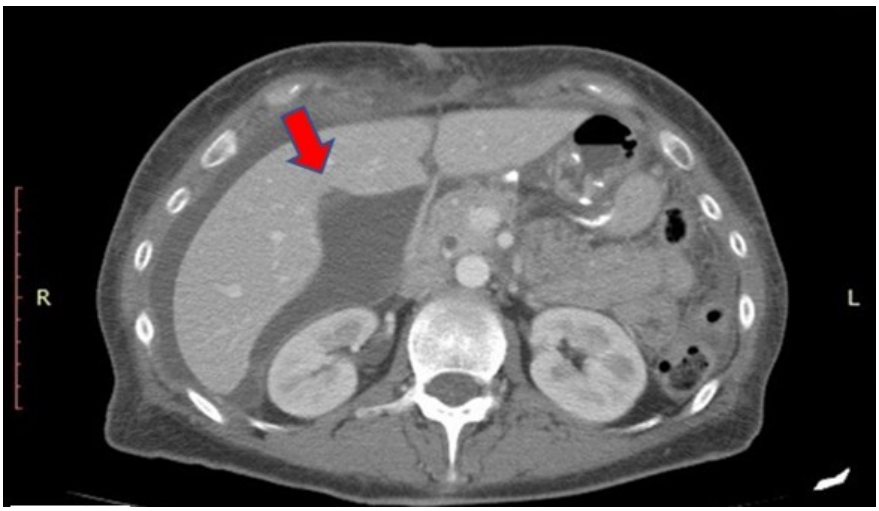

Figure 4. CT image of a Luschka duct leakage. Red Arrow indicates the bile collection.

Again, we would also like to emphasize the importance of the cooperation of a multidisciplinary team from different surgical units and subspecialties in the treatment of these complicated cases.

In cases of a Luschka leak after liver transplantation, ERCP is both diagnostic and therapeutic. Percutaneous transhepatic cholangiography (PTC) may be preferable to ERCP in the setting of Roux-en-Y Hepatic Jejunostomy given the altered anatomy. If ERCP or PTC fails, surgical treatment with repair is recommended. However, since the cause of a biliary leak after transplantation is often related to other major injuries, surgical exploration is the most preferred approach [64,65].

4.2. Limitations

The main limitation of our study is that it is a review of published articles of different regions and areas of expertise. Another point to note is the under-reporting of BDI in general and LD leaks. We recommend more reporting and future studies regarding these pathologies.

5. Conclusions

Leaks from LDs are rare. However, together with cystic stump leaks, they represent the second most frequent cause of bile leak after laparoscopic cholecystectomy. Thorough inspection of the gallbladder bed is recommended after an LC. Since these leaks are uncommon events, they tend to be described as sporadic case reports. A multidisciplinary team should approach and treat such cases.

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