Double cystic duct, a review of literature with report of a new case

Abdulwahid M. Salih a, F.H. Kakamad b,c,*, Shvan H. Mohammed c,d, Rawezh Q. Salih c,d, Imad J. Habibullah e, Aso S. Muhialdeen c,e, Hiwa Fatih f

a School of Medicine, Department of Sulaimani, Francois Mitterrand Street, Sulaimani, Kurdistan Region, Iraq
b School of Medicine, Department of Cardiothoracic and Vascular Surgery, University of Sulaimani, Francois Mitterrand Street, Sulaimani, Kurdistan Region, Iraq
c Bioscience Center, Hamdi Street, Sulaimani, Kurdistan Region, Iraq
d University of Sulaimani, Faculty of Science & Science Education, School of Science, Biology Department, Raperin Street, Sulaimani, Kurdistan Region, Iraq
e Sulaimani Teaching Hospital, Sulaimani, Kurdistan Region, Iraq
f School of Dentistry, University of Sulaimani, Francois Mitterrand Street, Sulaimani, Kurdistan Region, Iraq

A R T I C L E   I N F O

Article history:
Received 19 April 2017
Received in revised form 15 July 2017
Accepted 16 July 2017
Available online 21 July 2017

Keywords:
Double cystic duct
Double gallbladder
Common bile duct

A B S T R A C T

INTRODUCTION: Although cystic duct variation is quite common, duplication of cystic duct is an extreme rare variant. We report a case of double cystic duct with literature review. A 33-year-old female presented with right upper quadrant pain of three day duration, associated with nausea and poor appetite. The patient reported previous three attacks of right upper quadrant pain within the last two years. On examination: Murphy’s sign was positive and the right upper quadrant was tender. Abdominal ultrasound showed multiple gall stones. Oesophago-gastro-duodenoscopy was normal. Under general anesthesia, four port formal laparoscopy was done, double cystic duct was found. Histopathological examination showed features of chronic cholecystitis.

CONCLUSION: Double cystic duct is a very rare variant of the cystic duct anomaly. Identification pre or intraoperatively is very important to prevent ductal injury.

© 2017 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Although cystic duct variation is quite common, duplication of cystic duct is an extreme rare variant; only 16 cases have been reported in literature [1–15]. These variations increase risks of ductal injury, need for open conversion and postoperative complication [13]. The diagnosis is usually established during surgery [8]. However, it may be missed intraoperatively and diagnosed postoperatively during a diagnostic work up performed for persistent biliary symptoms [15].

In line with SCARE guide line, we report a case of a young female who had two separate cystic ducts exiting from a single gallbladder [16].

1.1. Patient information

A 33-year-old married female presented with right upper quadrant pain of three day duration. The pain started suddenly, sharp in character, awakening her from sleep, radiating to right shoulder and epigastric area, associated with nausea and poor appetite. The patient reported previous three attacks of right upper quadrant pain within the last two years that settled down within few days. Past surgical and medical histories were negative.

1.2. Clinical findings

the patient was in pain. Murphy’s sign was positive and right upper quadrant was tender.

1.3. Diagnostic assessment

Vital signs, blood examination, electrocardiography and echocardiography were within normal range. Abdominal ultrasound showed multiple gall stones, largest one about 14 millimeters in diameter. Oesophago-gastro-duodenoscopy (OGD) was normal.

1.4. Therapeutic intervention

Under general anesthesia, four port formal laparoscopy was done, double cystic duct was found (Fig. 1), clipped, gall bladder
Fig. 1. Intraoperative finding of double cystic duct. White arrows show the stumps of the first cystic duct. The blue arrow shows the second cystic duct prepared for division.

Fig. 2. Multiple stones evacuated from the gallbladder.

removed, and there were multiple gall stones (Fig. 2). Gross examination of the specimen confirmed double cystic duct (DCD) (Fig. 3). Histopathological examination showed features of chronic cholecystitis.

Fig. 3. Macroscopic examination of the specimen showing two separate openings in the gallbladder (the suture material).

1.5. Follow-up and outcomes

Two weeks later, the patient was stable and healthy.

2. Discussion

Presence of variations especially DCD during laparoscopic cholecystectomy is of great challenge for laparoscopic surgeons. Variation in the anatomy of cystic duct is not uncommon [7]. A classic anatomic picture between the extrahepatic bile duct, cystic duct, and related arteries is seen in only 33% of individuals. In 66% of the cases, the entry of the cystic duct into the common bile duct (CBD) is angular. In 17% to 20%, the CBD is parallel to the cystic duct and the entry is straighter. In 5% to 8%, the cystic duct is spiral and tortuous, connecting the CBD at a variable angle. In less than 1% of the patients, it drains into the right hepatic duct [8]. DCD is an extremely rare variant [16]. It is usually more common among female. From the reported cases, 11 cases (73%) occurred among female patients [3–11,14,15]. The age at the time of awareness of the variation varies ranging from new born to 76 years old [4,11]. Vincente et al. reported a newborn baby with VACTERL association (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities) having DCD discovered during cholecystectomy for symptomatic cholelithiasis [11]. The current case was a young, married female. Otaibi et al.

Table 1

| Reference, Authors | Year of publication | Age, sex | Diagnosis | Number of gallbladder | Presenting pathology |
|--------------------|---------------------|----------|-----------|-----------------------|----------------------|
| 1. Heyas et al.    | 1931                | 35. male | Preoperative CRE | double                | Not mentioned        |
| 2. Kannun         | 1933                | 69. male | Second re-exploration | double                | Suppurative cholecystis |
| 3. Wilson          | 1939                | 53. female | intraoperative | single | Postmortem educational examination |
| 4. Paraskevas et al | 1989              | 76. female | Postmortem | single | Chronic cholecystitis |
| 5. Nakasugi et al. | 1995                | 50. female | Preoperative ERCP | single | Chronic cholecystitis |
| 6. Hirono et al.   | 1997                | 74. female | Preoperative US, CT-scan | single | Chronic cholecystitis |
| 7. Tsutsumi et al. | 2000                | 74. female | Preoperative ERCP | single | Chronic cholecystitis |
| 8. Shishare et al. | 2002                | 46. female | intraoperative | single | Chronic cholecystitis |
| 9. Huston et al.   | 2006                | 43. female | intraoperative | single | Chronic cholecystitis |
| 10. Yoo et al.     | 2008                | 55. female | Preoperative MRI, PTBD | single | CBD cancer |
| 11. Vicente et al. | 2009                | newborn | Intraoperative | single | Acute cholecystitis |
| 12. Gürkem et al.  | 2014                | 10. male | Preoperative US | double | Acute cholecystitis |
| 13. Otaibi et al.  | 2015                | 54. male | intraoperative | single | Chronic cholecystitis |
| 14. Samanani et al.| 2015                | 43. female | intraoperative | single | Chronic cholecystitis |
| 15. Wei et al.     | 2015                | 66. female | Preoperative US, CT-scan | double | Chronic cholecystitis |

ERCP: endoscopic retrograde cholangiopancreatography; CRE: complete roentgen examination; MRI: magnetic resonance imaging; PTBD: percutaneous transhepatic biliary drainage; US: ultrasound; CT-scan: computed tomography scan; CBD: common bile duct.
stated that DCD associated with double gallbladder in 80% of the cases [13]. From the 15 reported cases in the literature, five cases (33%) associated with double gallbladder [1–3,12,15]. We did not find accessory gallbladder, the two cystic ducts drained from the same gallbladder. Flannery and Caster classified DCD into three different categories according the configuration of the ducts. The “H type” is the most common variant in which the second cystic duct drains separately into either the left, right, or common hepatic duct (CHD). The current patient had the Y type variant. In “Y type” variant, both cystic ducts unite to form a common channel that drains into the common hepatic duct. The third type variant is the trabecular type in which the accessory duct drains directly into substance of liver [8,14]. Preoperative diagnosis is difficult due to rarity of the variation and difficulty in visualization of the ducts by radiological imaging [2–4,11,14]. Ultrasound of the current case failed to reveal DCD. Some authors prefer routine use of intraoperative cholangiography to differentiate bile duct injury that might occur either because of transaction of bile duct or because of the presence of anatomical variations [7,14]. The limitation of this study is the absence of an intraoperative investigation like cholangiogram or post operative investigation like MRCP/ERCP to confirm the diagnosis. Table 1 shows literature review of the reported cases of DCD.

In conclusion, DCD is a very rare variant of the cystic duct variation. Identification pre or intraoperatively is very important to prevent ductal injury.

Consent
Informed consent was taken from the patient for the publication of this report.

Author contribution
Abdulwahid M. Salih: Surgeon performed the operation and follow up. F. H. Kakamad: writing the manuscript, reviewing the literature and follow up with final approval of the manuscript.
Rawezi Q.S, Shvan H.M, Imad J. Habibullah, Aso S. Muhiadeen, Hewa Fatih: major contribution to the idea, literature review and final approval of the manuscript.
Sources of funding
No source to be stated.

Ethical approval
Approval has been taken from bioscience centre.

Guarantor
Fahmi Hussein kakamad.

Conflicts of interest
There is no conflict to be declared.

References
[1] R. Hayes, Double gall bladder, with double cystic duct: case report. Radiology 16 (January) (1931) 66–67.
[2] R. Kennon, A double gall-bladder opening by two cystic ducts into the common bile-duct, Br. J. Surg. 20 (January 79) (1933) 522.
[3] C.L. Wilson, Double gallbladder with two cystic ducts and two cystic arteries, Ann. Surg. 110 (July 11) (1939) 60.
[4] G. Paraskevas, B. Papaziogas, K. Natsis, S. Spanidou, P. Kitsoulis, K. Atmatzidis, et al., An accessory double cystic duct with single gallbladder, Chirurgia (Bucharest, Romania) 102 (December 2) (2006) 223–225.
[5] H. Nakasuji, S. Kobayashi, K. Sakamoto, T. Inayoshi, M. Matsumura, M. Urabe, A case of double cystic duct with cholecystolithiasis treated by laparoscopy, J. Hepato-Biliary Pancreat. Surg. 2 (March 1) (1995) 68–71.
[6] Y. Hiroto, Y. Takita, N. Nitta, H. Hashimoto, Double cystic duct found by intraoperative cholangiography in laparoscopic cholecystectomy, Surg. Laparosc. Endosc. Percutanous Tech. 1 (June 3) (1997) 263–265.
[7] S. Tsutsumi, Y. Hosouchi, T. Shimura, T. Asao, T. Kojima, S.I. Takenoshita, et al., Double cystic duct detected by endoscopic retrograde cholangiopancreatography and confirmed by intraoperative cholangiography in laparoscopic cholecystectomy: a case report, Hepatogastroenterology 47 (December 35) (1999) 1266–1268.
[8] R. Shivhare, S.S. Sikora, Double cystic duct: a rare biliary anomaly encountered at laparoscopic cholecystectomy, J. Laparoendosc. Adv. Surg. Tech. 12 (October 5) (2002) 391–392.
[9] T.L. Huston, G.F. Dakin, Double cystic duct, Can. J. Surg. 51 (February 1) (2008) E9.
[10] J. Yoo, D. Shin, An accessory double cystic duct with a single gall bladder, J. Korean Surg. Soc. 74 (April 4) (2008) 319–321.
[11] H. Lugo-Vicente, M. Correa, H. Brunet, Double duct in a child withvacterl association: a case report, Boletin de la AsociacionMedica de Puerto Rico 101 (2) (2009) 56.
[12] S.B. Gökrem, S. Doğanay, G. Kahirman, M. Küçükaydın, A. Koçun, Acute cholecystitis of a duplicated gallbladder with double cystic duct in a 10 year old boy, Balkan Med. J. 31 (December 4) (2014) 366.
[13] W. Otabi, G. Quach, B. Burke, Double cystic duct in a septated gallbladder, J. Investigit. Med. High Impact Case Rep. 3 (April 2) (2015) 321–323.
[14] S.S. Samnani, A. Ali, Y variant of double cystic duct: incidental finding during laparoscopic cholecystectomy, Indian J. Surg. 77 (December Suppl 3) (2015) 1491.
[15] W. Yu, H. Yuan, S. Cheng, F. Xing, W. Yan, A double gallbladder with a common bile duct stone treated by laparoscopy accompanied by cholecdochoscopy via the cystic duct: a case report, Exp. Ther. Med. 12 (December 6) (2016) 3521–3526.
[16] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, Orgill DP and the SCARE group: the SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.