An unusual variation of gallbladder duplication originating from the right hepatic duct

Christine Wong a, Sonia Mason a, David Bowden c, Rebecca Brais d, Simon Harper b,*

a School of Clinical Medicine, Addenbrooke’s Hospital, Hills Rd, Cambridge, CB2 0SP, UK
b Department of Surgery, Addenbrooke’s Hospital, Hills Rd, Cambridge, CB2 0SP, UK
c Department of Radiology, Addenbrooke’s Hospital, Hills Rd, Cambridge, CB2 0SP, UK
d Department of Pathology, Addenbrooke’s Hospital, Hills Rd, Cambridge, CB2 0SP, UK

ABSTRACT

INTRODUCTION: Duplication of the gallbladder is a rare congenital biliary anomaly and may present with similar pathology to that seen in a single gallbladder. We present a previously unreported case of a symptomatic duplex gallbladder arising directly from a long segment of the right hepatic duct.

PRESENTATION: A 23 years old female was referred to our team with right upper quadrant pain suggestive of biliary colic. Ultrasound, contrast enhanced CT and magnetic resonance cholangiopancreatography revealed a normal gallbladder and a separate cystic lesion containing multiple gallstones and communicating with the right main hepatic duct. Surgical management involved cholecystectomy, resection of the cystic lesion from the right hepatic duct and reconstruction with hepaticojejunostomy. The patient made a good recovery from surgery, reporting complete resolution of symptoms. Histology of the cystic lesion confirmed duplicate gallbladder with features of severe chronic cholecystitis.

DISCUSSION: Symptomatic duplicate gallbladders warrant cholecystectomy and in more straightforward variations, in which a shared, single cystic duct is encountered, a laparoscopic approach is feasible. Surgical management may be more complicated for anomalies in which the duplicate gallbladder is connected separately and more proximally in the biliary tree.

CONCLUSION: Trabecular duplicate gallbladder, in which a second gallbladder originates from the right or left hepatic duct is extremely rare. We report a previously undescribed variation, in which the gallbladder is attached over a wide area to the right hepatic duct and outline the successful surgical management undertaken.

© 2018 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

Duplication of the gallbladder is a rare congenital biliary anomaly with a reported incidence of 1 in 4000 [1]. Duplicate gallbladders may present with diseases similar to those occurring in a single gallbladder and management options include both open and laparoscopic cholecystectomy [2]. Herein, we present a previously unreported case of a symptomatic duplex gallbladder arising directly from a long segment of right hepatic duct, in the absence of a connecting cystic duct. This case report has been reported in line with the SCARE criteria [3].

2. Presentation of the case

A 23 years old female was referred to our department, presenting with a 2-month history of right upper quadrant pain, radiating to the shoulder and back, occurring after meals, and highly suggestive of biliary colic. The patient was otherwise fit and well and had no significant past medical history. On further investigation she was found to have mildly deranged liver function tests (LFT). Imaging with ultrasound scan (US) and contrast enhanced computerised tomography (ceCT) demonstrated a 45 mm cystic lesion in the liver hilum (Fig. 1).

The liver lesion was further delineated using magnetic resonance cholangiopancreatography (MRCP) and this showed an unremarkable, normally positioned gallbladder along with a separate cystic lesion, which contained multiple gallstones and communicated directly with the right main hepatic duct (Fig. 2).

These findings were felt to be most consistent with a differential diagnosis of gallbladder duplication, intraductal papillary mucinous neoplasm – biliary type or intra-hepatic choledochal cyst.
Clinically, the diseases of duplicate gallbladder are similar to those associated with single gallbladder, including cholecystocholangitis, cholecystitis, cholelithiasis, cholangitis and carcinoma [9]. However, there is no evidence to show that this anomaly in itself is associated with higher risk of cholecystitis or malignancy, and so prophylactic cholecystectomy in an asymptomatic patient with gallbladder duplication is not recommended [10]. It is common for duplication of gallbladder to remain undetected prior to cholecystectomy [11] and this is likely to carry a higher risk of intra-operative biliary and vascular injury. Pre-operative recognition of double or multiple gallbladders is also preferable due to the high incidence of persistent symptoms and complications should a duplicate gallbladder be left behind, usually necessitating further surgery [8,12,13]. It is also important to consider the differential diagnoses for gallbladder duplication prior to resection, which include a gallbladder with a large floppy fundus, gallbladder diverticula, gallbladder fold, Phrygian cap, choledochal cyst, pericholecystic fluid, focal adenomyomatosis and intraperitoneal fibrous bands [14].

A clear diagnosis and surgical planning typically requires multi-modal imaging comprising USS, ceCT and MRCP. Minimally invasive approaches, including single incision laparoscopic and robotic cholecystectomy are widely reported for gallbladder duplication, but are generally reserved for Type 1 split gallbladders [2,15,16]. Although successful laparoscopic procedures have also been described for Type 2 gallbladder duplication [17], on the whole, open surgery is advocated due to the added complexity of surgery relating to intrahepatic position of the gallbladder and high insertion of the second cystic duct. The use of intra-operative cholangiography is recommended to confirm the diagnosis of duplication and aid surgical planning [8].

4. Conclusion

In our case, the patient was found to have a Type 2, right trabecular accessory gallbladder. Trabecular accessory gallbladders are by far the rarest subtype, accounting for only 4 cases in a series of 148 [18] patients with gallbladder duplication and there are very few reports describing an accessory gallbladder arising from the right hepatic duct [18,19]. A further unusual and previously undescribed feature in this case was the absence of a narrow cystic duct, with direct communication between the wide gallbladder neck and the right hepatic duct. This variation was successfully managed however, with multimodal imaging for surgical planning and reconstruction of the right hepatic duct drainage via hepaticojejunostomy formation.

Conflicts of interest

None.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

The report was deemed exempt from formal ethical approval by the Author’s institution.

Consent

Patient’s written consent was obtained.

Fig. 1. Unenhanced CT image showing a well demarcated fluid filled mass within the gallbladder fossa and separate from the gallbladder. No evidence of calcification.
Fig. 2. (a) Axial T2-weighted MR image demonstrating a fluid and calculus-filled cystic mass within the liver hilum and possibly segments 4b/5 of the liver parenchyma. (b) Coronal maximum intensity projection (MIP) MRCP image illustrates communication between the extrahepatic gallbladder and CBD via the cystic duct. Calculi and biliary sludge is seen within the dilated segment 8 bile ducts. No cystic duct from the intrahepatic gallbladder could be visualised. (c) Cinematic volume rendered technique (VRT) images using magnetic resonance cholangiopancreatography (MRCP) data, which highlights fluid-filled structures. The presence of multiple filling defects within the intrahepatic gallbladder results in the impression of a lobulated mass.
The statement ‘Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor−in−Chief of this journal on request’ was added to the end of the manuscript.

Author contribution
Christine Wong – Writing of Manuscript.
Sonia Mason - Writing of Manuscript.
Dr Dave Bowden – Providing radiological images, writing of manuscript.
Dr Rebecca Brais – Writing of manuscript.
Mr Simon Harper – Identification of case, Writing of manuscript.

Registration of research studies
N/A.

Guarantor
Mr Simon Harper.

Provenance and peer review
Not commissioned, externally peer-reviewed.

Acknowledgements
None.

References
[1] F.A. Boyden, The accessory gall-bladder− an embryological and comparative study of aberrant biliary vesicles occurring in man and the domestic mammals, Am. J. Anat. 38 (2) (1926) 177−231.
[2] A. Al Rawahi, Y. Al Azri, S. Al Jabri, A. Alfadhi, S. Al Aghbari, Successful laparoscopic management of duplicate gallbladder: a case report and review of literature, Int. J. Surg. Case Rep. 21 (2016) 142−146.
[3] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, for the SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180−186.
[4] M. Lamah, N.D. Karanjia, G.H. Dickson, Anatomical variations of the extrahepatic biliary tree: review of the world literature, Clin. Anat. 14 (2001) 167−172.
[5] M.W. Causey, S. Miller, C.A. FERNELIUS, J.R. Burgess, T.A. Brown, C. Newton, Gallbladder duplication: evaluation, treatment, and classification, J. Pediatr. Surg. 45 (2) (2010) 443−446.
[6] R. Gross, Congenital anomalies of the gall bladder: a review of one hundred and forty-eight cases, with report of a double gall bladder, Arch. Surg. 32 (1936) 131.
[7] N. Harlaftis, S.W. Gray, J.E. Skandalakis, Multiple gallbladders, Surg. Gynecol. Obstet. 145 (6) (1977) 928−934.
[8] B.D. Kim, I. Zendejas, C. Velopulos, S. Fujita, J.F. Magliocca, L.K. Kayler, A.W. Hemming, Duplicate gallbladder arising from the left hepatic duct: report of a case, Surg. Today 39 (6) (2000) 536−539.
[9] W.J. Roeder, W.L. Mersheimer, K.K. Kazarian, Triplication of the gallbladderwith cholecystitis, cholelithiasis and papillary adenocarcinoma, Am. J. Surg. 121 (1971) 746−748.
[10] Y. Pillay, Gallbladder duplication, Int. J. Surg. Case Rep. 11 (2015) 18−20.
[11] M. Hishinuma, Y. Isogai, Y. Matsuura, M. Kodaira, S. Oi, N. Ichikawa, et al., Double gallbladder, J. Gastroenterol. Hepatol. 19 (2004) 233−235.
[12] R. Silvis, A.J. van Wieringen, C.H. van der Werken, Reoperation for a symptomatic double gallbladder, Surg. Endosc. 10 (1996) 336–337.

[13] F. Borghi, G. Giraudo, P. Geretto, et al., Perforation of missed double gallbladder after primary laparoscopic cholecystectomy: endoscopic and laparoscopic management, J. Laparoendosc. Adv. Surg. Tech. A 18 (2008) 429–431.

[14] J. Gigot, B. Van Beers, L. Goncette, et al., Laparoscopic treatment of gallbladder duplication. A plea for removal of both gallbladders, Surg. Endosc. 11 (5) (1997) 479–482.

[15] M.A. Boyle, A.W. Kaplin, L. Kushnir, P. Montero-Pearson, J. Robot. Surg. 10 (2) (2016) 161–163.

[16] E. Gürbulak, H. Özşahin, Y. Düzköylü, I. Akgün, M. Battal, B. Gürbulak, Single incision laparoscopic cholecystectomy for gallbladder duplication, Case Rep. Surg. (2015), e589313.

[17] J.M. Maddox, M.L. Demers, Laparoscopic management of gallbladder duplication: a case report and review of literature, JSLS 3 (2) (1999) 137–140, 1999.

[18] B. Singh, L. Ramsaroop, L. Allopi, J. Moodley, K. Satyapal, Duplicate gallbladder: an unusual case report, Surg. Radiol. Anat. 28 (2006) 654–657.

[19] D. Weibel, M. Kaufmann, H.J. Riedtmann-Klee, Accessory gallbladder originating from the right hepatic duct, Surg. Endosc. 15 (5) (2001) 519.