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ORIGINAL ARTICLE

Focal Intrahepatic Duct Dilatation (FIDD): a Finding That Mandates Further Evaluation That May Amount to Liver Resection

Nagappan Kumar[1] · Arvind Kumar[2] · Dipanjali Mondal[3]

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Abstract

The increasing use of imaging has led to incidental findings in the liver. The Western experience of managing focal intrahepatic duct dilatation (FIDD) is not well recorded. We present our experience based on a large prospectively maintained database at a tertiary hepatobiliary surgical unit. We identified patients with liver resection for focal incidental duct dilatation from January 2003 to December 2019 from the liver unit database. We recorded the demographics, symptomatology, blood test results, imaging, type of liver resection, morbidity, mortality and histology of resected specimens. Nine patients had focal intrahepatic duct dilatation among 994 liver resections performed (0.9%). Six patients were asymptomatic, 2 upper abdominal pain and 1 recurrent gram-negative sepsis. Liver function tests were normal in all patients. Two patients had cholangiocarcinoma (CCA), 4 intrahepatic stones, 1 intraductal papillary neoplasm of bile duct (IPN-B) and 2 benign strictures. Focal incidental duct dilatation is rare in the Western population. Most patients are asymptomatic with an incidental finding of intrahepatic duct dilatation on cross-sectional imaging. Differentiating benign and malignant pathology is difficult warranting liver resection, in fit patients, to resolve the diagnosis. Liver resection is safe and can be potentially curative in patients with a neoplasm, which can occur in 30% of patients with focal intrahepatic duct dilatation.

Keywords Bile duct · Intrahepatic · Dilatation · Cholangiocarcinoma · Hepatectomy

Introduction

Advances in diagnostic imaging have resulted in an escalation in cross-sectional imaging in the modern era. The increasing use of computed tomography (CT) and ultrasound scanning in assessing acutely unwell patients to evaluate organs such as the colon, renal tract and lungs has resulted in a rise in the detection of patients with incidentally identified bile duct dilatation [1, 2]. Bile duct dilatation is not an uncommon finding in completely asymptomatic patients or in patients with vague abdominal pain. The most commonly studied duct has been the extrahepatic common bile duct (CBD). Multiple studies have recommended follow-up investigation for incidentally identified CBD dilatation to exclude serious underlying aetiology [3–5]. However, there has been very little published literature regarding the clinical significance and outcomes of patients with incidental FIDD [6].

We present a case series of nine patients in whom FIDD was incidentally identified and who then underwent liver resection to obtain a definitive diagnosis. Histology confirmed a treatable pathology in the majority of cases, including curable malignancy. Our experience suggests that these incidental findings on radiological examinations should be investigated further and that they may well require liver resection to resolve the diagnosis.

✉ Nagappan Kumar
Nagappan.kumar@wales.nhs.uk

Arvind Kumar
a.kumar5@nhs.net

Dipanjali Mondal
dipanjali.mondal@wales.nhs.uk

[1] HPB Surgery, Cardiff Liver Unit, University Hospital of Wales, Cardiff CF14 4XW, UK

[2] Croydon University Hospital, Surrey CR7 7YE, UK

[3] Department of Radiology, University Hospital of Wales, Cardiff CF14 4XW, UK

Methods

We identified patients with FIDD from a prospectively maintained database at a tertiary regional liver surgery unit, from January 2003 to December 2019. At referral, all patients had a contrast enhanced CT (CECT) scan of the abdomen and pelvis where the FIDD was identified. We obtained further history from each patient and performed liver function tests (LFTs), CA19-9 and MRI/MRCP. One patient had a peroral Spyglass™ cholangioscopy. We did not attempt *peroral cholangioscopy in others because either they were seen in the earlier years of the series when this procedure was not available or the intrahepatic duct dilatation was beyond second-order ducts and hence beyond the reach of the cholangioscopy*. We performed laparoscopy and CT chest on all patients to exclude peritoneal metastases and lung metastases, respectively, since cholangiocarcinoma (CCA) was always within the differential diagnosis for FIDD. *The unit's policy was to offer liver resection to all patients with FIDD even when the aetiology remained unclear after all possible investigations*. We recorded the demographics, preoperative imaging, LFTs, CA19-9, operative details including type of procedure, blood loss, blood transfusion, postoperative morbidity using Clavien-Dindo classification [7], hospital stay, mortality and histology of the resected specimen. We defined liver resection as major if it involved three or more segments [8]. All the available radiological examinations were retrospectively reviewed by one fellowship trained radiologist (DM), with 7 years subspecialty experience in hepatopancreaticobiliary imaging, to ascertain whether a definitive diagnosis could have been made preoperatively. All patients were followed up prospectively and data censored at death or 30 June 2020. Formal ethics committee approval was not deemed necessary, since the study was regarded as retrospective notes review.

Results

We performed 994 liver resections between January 2003 and December 2019, with 9 (0.9%) performed for FIDD. All patients investigated for FIDD during the study period underwent surgery; thus, the current study included all patients with FIDD referred to our unit. The median age was 64 (range 52–82 years) with 4 men and 5 women.

The demographics, presenting symptoms, brief description of radiological findings, LFTs and CA19-9 are given in Table 1. The operative details including type of resection, blood loss, units of blood transfused, hospital stay, morbidity, mortality and final histology is given in Table 2.

Two patients had a histological diagnosis of CCA (an example is shown in Fig. 1), four had intrahepatic biliary stones (an example shown in Fig. 2), one patient had intraductal papillary neoplasm of the bile duct (IPN-B) (see Fig. 3) and two patients had benign strictures.

We followed up patients for a median of 5 years (1–16 years). Two patients with CCA and one with intrahepatic stone disease died during follow up. An 82-year-old gentleman who had a left hepatectomy for CCA developed a solitary recurrence in segment 8 of the liver 42 months later, which was treated with microwave ablation. He developed further recurrence at the edge of the ablation zone and died 6 years after the initial surgery. An 81-year-old lady with CCA who had a right hepatectomy died 3 years after surgery from a new oesophageal carcinoma, with imaging showing no evidence of recurrent CCA. The patient who had intrahepatic stone died 8 years after surgery due to an unrelated cause.

Table 1 Imaging, biochemistry (liver function test [LFT]), clinical symptom and histology for all patients with FIDD

Patient	Age/gender	Clinical symptoms	Radiological finding	LFT	CA 19–9	Clinical symptoms
1	61/M	RUQ pain	Focal dilatation proximal to left main hepatic duct	Normal	51	RUQ pain
2	82/M	None	Dilatation of ducts in left lateral segment	Normal	Not done	None
3	64/F	Epigastric pain	Dilated left hepatic duct	ALP 181	Normal	Epigastric pain
4	81/F	None	Intrahepatic duct dilatation with atrophy of segments 6 + 7	Normal	96	None
5	78/F	RUQ pain	Focal dilatation in segments 2&3	Normal	Normal	RUQ pain
6	52/F	None	Focal biliary dilatation in segments 6&7	Normal	Normal	None
7	55/M	None	Isolated duct dilatation in segment 7	Normal	Normal	None
8	58/F	None	Isolated duct dilatation in segment 6&7	Normal	Normal	None
9	74/M	None	Dilatation of ducts in segment 8	Normal	Normal	None

ALP alkaline phosphatase, *RUQ* right upper quadrant

Table 2 Operation performed and postoperative morbidity and in hospital mortality for patients with FIDD

Patient	Type of resection	Blood loss (mls)	Units of blood transfused	Length of hospital stay (days)	Morbidity (Clavien Dindo classification)	In hospital mortality	Histology
1	Left lateral sectionectomy	100	0	15	None	None	Hepatolithiasis
2	Left hepatectomy	520	0	8	Chest infection (CD2)	None	CCA
3	Left hepatectomy	200	0	7	None	None	Hepatolithiasis
4	Right hepatectomy	600	0	21	Subphrenic collection requiring drainage (CD3)	None	CCA
5	Left hepatectomy	100	0	8	None	None	IPN-B
6	Right hepatectomy	150	0	5	None	None	Benign stricture
7	Right hepatectomy	800	0	4	None	None	Hepatolithiasis
8	Right hepatectomy	300	0	12	Chest infection (CD2)	None	Benign stricture
9	Right anterior sectionectomy	1740	2	5	None	None	Hepatolithiasis

CCA cholangiocarcinoma, *IPN-B* intraductal papillary neoplasm-bile duct

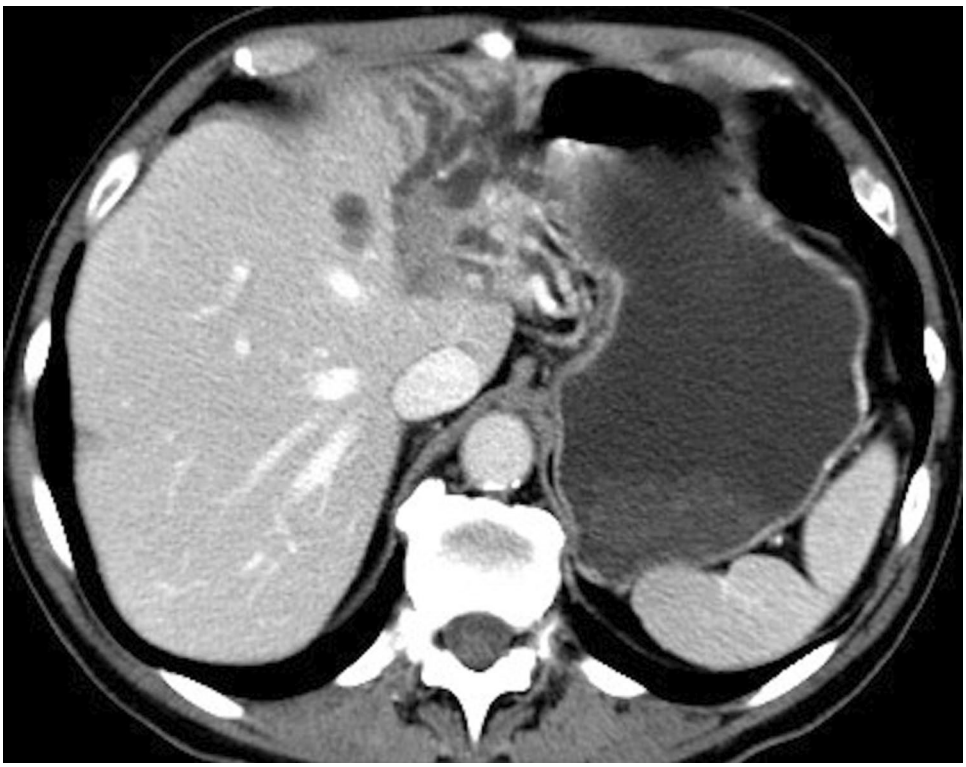

Fig. 1 CT axial image demonstrating infiltrating cholangiocarcinoma causing biliary obstruction and resultant atrophy in the left lobe of the liver

Discussion

The identification of radiological abnormalities unrelated to the patient's presenting complaint is increasing due to the more widespread use of diagnostic cross-sectional imaging. A white paper on incidental radiological findings, including in the gallbladder and biliary tree, by the American College of Radiologists, focuses on the extrahepatic duct but not intrahepatic biliary tree [9]. The management of incidental findings on abdominal CT including CT colonography has been previously described; however, these were mostly focused on incidental lesions (e.g., liver, renal and adrenal lesions); there has been no guidance for the management of FIDD [2]. A study described [10] described 24 patients with intrahepatic bile duct dilatations, but many patients had jaundice and were therefore not obviously incidental findings. This study defined intrahepatic duct dilatation as diameter more than 0.8 cm for

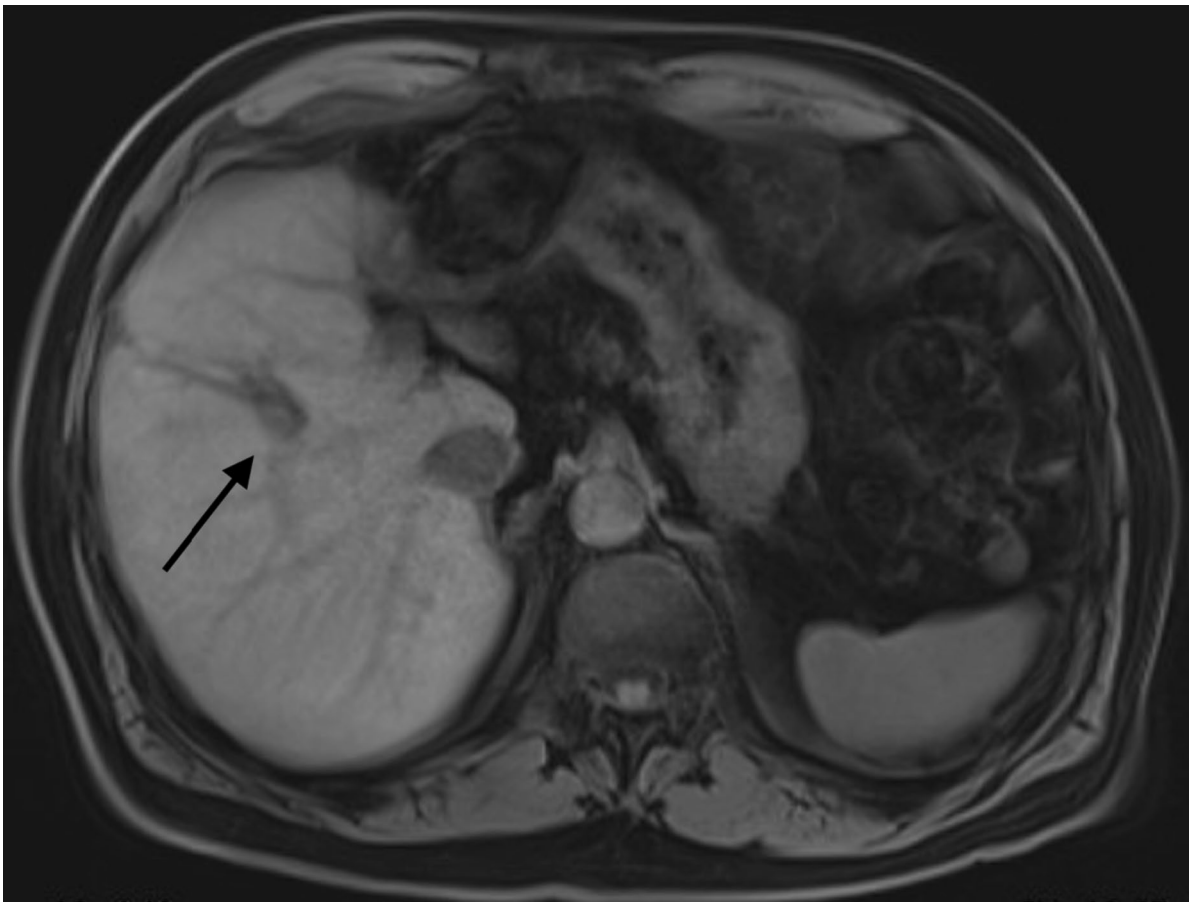

Fig. 2 MRI T1 axial image showing high T1 signal intraductal calculus (arrow)

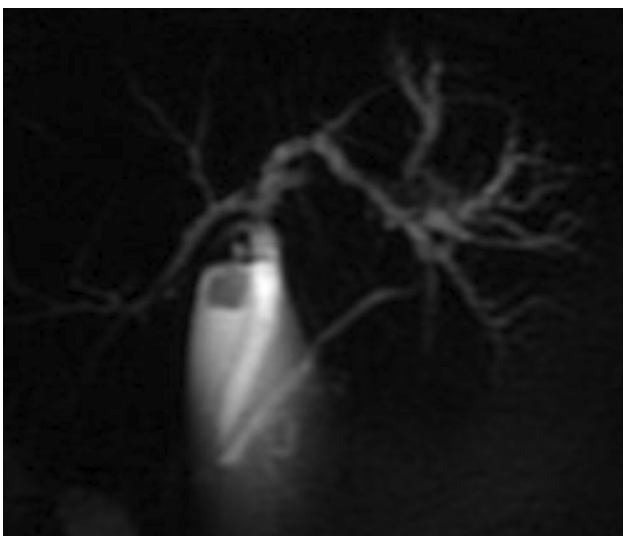

Fig. 3 MRCP image shows predominantly left sided and segments 8 and 5 intrahepatic duct dilatation. No cause for this was demonstrated on imaging. Histopathology confirmed IPN-B

the right hepatic duct, more than 1 cm for the left hepatic duct and more than 0.5 cm for more peripheral ducts.

The optimal strategy to determine which patient with radiologically confirmed FIDD warrants further investigation remains uncertain. The presumed diagnosis in all patients is a small intrahepatic CCA which is too small to cause mass effect, but enough to cause biliary obstruction and proximal dilatation. The previously quoted study found that serum alkaline phosphatase and CA19-9 were significantly more elevated in patients with malignant pathology than in benign pathology [10]. Another study showed that CA19-9 of more than 100 U/ml could identify patients with malignant pathology [11]. However, significant elevation of CA19-9 was not found in any of our patients where FIDD was detected incidentally; hence, tumour markers and liver function tests may not be helpful in distinguishing benign from malignant pathology.

Retrospective review of the radiological investigations identified intrahepatic stones in one patient in our series, as shown in Fig. 2. This may have been potentially managed with percutaneous transhepatic cholangioscopy and the stones retrieved. Dilatation of the stricture has also been

described as an alternative treatment strategy [12]. Ductal dilatation and stenting make routine hepatectomy unnecessary for left hepatolithiasis with intrahepatic biliary stricture [12], though not currently available at our centre. Percutaneous transhepatic cholangioscopy has been described to directly visualise intrahepatic biliary strictures [13]. Another study reported superior clarity of narrow band imaging (NBI) compared to white light imaging in identifying proximal tumour margins allowing better surgical planning; however, this was based on a small case series [13]. These techniques have yet to be widely adopted in the Western world. The limitations of percutaneous cholangioscopy are potential biliary fistula, cholangitis and the risk of tumour seeding. Peroral cholangioscopy (SpyGlass™) was used in one patient, but the site of stricture in the segment 6 duct could not be reached for formal assessment.

Our experience suggests that a finding of incidental FIDD is uncommon in the Western world (0.9%). *This could be an underestimate as patients who have not been operated were not included in this series. However, liver resection was offered and taken up by all patients to whom it was offered. Patients did not have surgery only if they were deemed unfit to undergo surgery and were not followed up*. The cases here highlight some of the aetiologies for incidental FIDD. We further discuss the significance of CCA, intrahepatic ductal calculi and intraductal papillary mucinous neoplasm of bile ducts (IPMN-B).

CCA is an aggressive, malignant adenocarcinoma affecting the bile ducts [14, 15]. Three recognised subtypes are described: intrahepatic, perihilar and distal [16]. The intrahepatic subtype is less common accounting for 25% of all CCA [15]. Together, they are the second most common primary hepatic malignancy [17] following hepatocellular carcinoma. All types of CCA have a poor prognosis with a 5-year survival for locally advanced extrahepatic (peri-hilar and distal) CCA at around 30% [18]. The poor prognosis for CCA is due to many factors including, but not limited to, late manifestation of symptoms within the natural history of the disease and the fact that many patients do not present with recognised risk factors.

In our study, we present two patients who were subsequently found to have intrahepatic CCA, thus suggesting that even in asymptomatic patients with normal liver function, further investigation and liver resection should be considered when FIDD is identified. The outcomes have been good with one patient surviving 6 years after surgery. The second patient died of a different unrelated oesophageal cancer with no evidence of CCA recurrence. The good outcomes demonstrated in these patients are a combination of early detection and therefore management, as well as favourable tumour biology.

One patient had intraductal papillary neoplasm of the bile duct (IPN-B). This is a relatively new diagnostic entity, which has arisen from a World Health Organisation (WHO) reclassification of tumours [19–21] IPN-B shows a spectrum that ranges from adenoma to adenocarcinoma, similar to colonic polyps with dysplasia progressing to carcinoma. Four types have been described: papillary, cast-like intraductal, superficial spreading and cystic [22].

Following resection, histopathological confirmation has demonstrated that as high as 40–94% of IPN-B has foci of invasive carcinoma [22, 23]. Even with transformation to CCA, IPN-B has a better 5-year survival than conventional CCA with a median survival of 52 months compared with 28 months [24]. The single patient in our series with a confirmed IPN-B had a papillary lesion with mucin production but no evidence of high-grade dysplasia or invasive malignancy. This was a premalignant lesion, and following resection, the patient is alive and disease free at 6 years of follow up.

We found hepatolithiasis in four patients with FIDD. In two patients, there was evidence of chronic inflammation secondary to the hepatolithiasis. Despite this histological evidence, the liver function was either within normal range or, in the case of one patient, an ALP of 180 (normal range 30–159) was the only abnormality. Evidence suggests that hepatolithiasis can lead to an increased risk of intrahepatic CCA [25] as well as pyogenic cholangitis [26]. The first patient in this series presented with recurrent *Escherichia coli* sepsis that persisted after clearing all the CBD stones. The liver abnormality was the only potential source of sepsis; therefore, surgery was performed to remove the left lateral segment which showed dilated ducts with resultant atrophy.

A challenge presented by these results is that, for many patients with incidentally identified FIDD, further management is either close monitoring or a major operation. Due to the paucity of available literature regarding the causes of FIDD, it can be difficult to justify major operation in asymptomatic patients as the treatment carries a potential mortality of 2–4% [27]. The patients in this series were all counselled pre-operatively and informed that they were to undergo a major surgical procedure for an asymptomatic pathology and gave informed consent. The rarity of the condition also does not allow development of evidence-based guidelines by which surgical teams can decide on whether an operative intervention is indicated. Modern techniques like percutaneous transhepatic cholangioscopy may allow non-operative treatment in a small proportion of patients, although this is currently not widely available.

There are limitations to this study. This is a small series that does not allow any statistical evaluation of predictive factors and cutoff values for tumour markers and LFTs that may predict malignancy. However, even benign conditions like intrahepatic stones need treatment. *The other limitation is the possible selection bias where only patients who had*

liver resection have been included. Although the policy of the unit was to resect all patients referred for intrahepatic duct dilatation, some patients may have chosen not to have resection and may not be included. However, all patients eligible for resection were operated, and none were denied surgery due to anatomical considerations. The strengths of this study are the long follow-up and good outcomes in the treated patients.

We conclude, based on our experience, that patients with focal intrahepatic duct dilatation need further investigation as 80% have a significant pathology that will lead to potential problems in the future and 30% could have cholangiocarcinoma that may not be diagnosed by any existing investigative modality. Liver resection may be the only method by which we can diagnose and treat this condition.

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