Clinical Journal of Gastroenterology (2023) 16:599–604
https://doi.org/10.1007/s12328-023-01801-1

CASE REPORT

Use of endoscopic gallbladder stenting and biliary scintigraphy for diagnosis of gallbladder dyskinesia: a case report

Masahiro Ono[1] · Hideki Kamada[1] · Kiyoyuki Kobayashi[2] · Toshiaki Kono[1] · Daisuke Namima[1] · Naoki Fujita[1] · Hiroki Yamana[1] · Joji Tani[1] · Hideki Kobara[1] · Tsutomu Masaki[1]

Received: 9 February 2023 / Accepted: 3 April 2023 / Published online: 12 April 2023

Abstract

A 40-year-old woman visited our hospital with a several-year history of right hypochondriac pain and vomiting after eating. She had been treated for functional dyspepsia, with no improvement in her symptoms. No gallstones were detected on imaging tests, but papillary insufficiency or dyskinesia of the gallbladder was suspected and biliary scintigraphy was performed. Biliary scintigraphy showed delayed excretion of radionuclides from the gallbladder and bile ducts into the duodenum. We initially suspected papillary dysfunction and performed endoscopic sphincterotomy, but there was no improvement in her symptoms. Biliary scintigraphy also showed delayed excretion of radionuclides, especially stagnation of radionuclides in the gallbladder. We suspected gallbladder dyskinesia and performed endoscopic gallbladder stenting, after which her symptoms disappeared and biliary scintigraphy showed improved excretion of radionuclides into the duodenum. Endoscopic gallbladder stenting may be useful for the diagnosis of gallbladder dyskinesia and for determining the efficacy of cholecystectomy.

Keywords Gallbladder dyskinesia · Endoscopic gallbladder stenting · Biliary scintigraphy · Cholecystectomy

Introduction

Dyskinesia of the gallbladder causes gallstone-like symptoms including abdominal pain and vomiting as a result of functional abnormalities of bile flow, in the absence of any organic abnormalities in the biliary system, including the gallbladder. Cholecystectomy is a common treatment, but it is often difficult to diagnose because its clinical symptoms resemble those of irritable bowel syndrome and some psychogenic disorders.

Here, we report a case of biliary dyskinesia in which biliary scintigraphy performed before and after endoscopic gallbladder stenting (EGBS) was predictive of successful cholecystectomy.

✉ Masahiro Ono
ono.masahiro@kagawa-u.ac.jp

[1] Department of Gastroenterology and Neurology, Faculty of Medicine, Graduate School of Medicine, Kagawa University, 1750-1 Ikenobe, Miki-cho, Kita-gun, Kagawa 761-0793, Japan

[2] Division of Innovative Medicine for Hepatobiliary and Pancreatology, Kagawa University, Kagawa 761-0793, Japan

Case report

A 40-year-old woman presented with a several-year history of epigastric and right quadrant pain and vomiting after eating. She had been treated for functional dyspepsia but her symptoms did not improve, and she was, therefore, referred to our hospital for a thorough examination.

Her height was 158.2 cm, weight was 53.1 kg, and her body mass index was 21.2 kg/m^2. She had a history of nephrotic syndrome, which was diagnosed when she became pregnant, and was in remission on 3 mg prednisolone at the time of examination. Blood tests showed free T3 2.18 U/ml, which was slightly below the normal range, but otherwise within normal limits (Table 1).

During a medical interview, she complained of pain in the right quadrant, especially after lipid intake, and biliary pain was suspected. Abdominal plain computed tomography showed no organic disease of the biliary system, such as gallstones or common bile duct stones (Fig. 1).

Endoscopic ultrasound also revealed no gallstones or stenosis, including debris in the gallbladder or bile ducts (Fig. 2).

In light of the patient's biliary pain but absence of organic disease, we suspected a functional gallbladder or Oddi's

Table 1 Laboratory findings at first medical examination

Hematology		TP	7.0 g/dl
WBC	5950/μl	Alb	4.6 g/dl
RBC	414×10^4/μl	BUN	11.7 mg/dl
Hb	12.8 g/dl	Cre	0.64 mg/dl
Ht	38.5%	Na	141 mmol/l
Plt	18.2×10^4/μl	K	3.7 mmol/l
		Cl	107 mmol/l
Biochemistry		Ca	9.4 mg/dl
T-Bil	0.8 mg/dl		
D-Bil	0.1 mg/dl	Serology	
GOT	18 IU/l	CRP	0.01 mg/dl
GPT	10 IU/l	TSH	0.478 μg/ml
ALP	99 IU/l	FT3	2.18 U/ml
γ-GTP	10 IU/l	FT4	1.09 U/ml
LDH	209 IU/l	HbA1c	5.4%

WBC white blood cell count, *RBC* red blood cell count, *Hb* hemoglobin, *Ht* hematocrit, *Plt* platelets, *T-Bil* total bilirubin, *D-Bil* direct bilirubin, *GOT* glutamic-oxaloacetic transaminase, *GPT* glutamic pyruvic transaminase, *ALP* alkaline phosphatase, *γ-GTP* gamma-glutamyl transferase, *LDH* lactate dehydrogenase, *TP* total protein, *Alb* albumin, *BUN* blood urea nitrogen, *Cre* creatinine, *CRP* C-reactive protein, *TSH* thyroid stimulating hormone, *FT3* free triiodothyronine, *FT4* free thyroxine

sphincter disorder, according to the Rome IV criteria, and performed biliary scintigraphy for supplementary diagnostic purposes (Fig. 3).

Biliary scintigraphy shows that the accumulation of radionuclides increases with time without decreasing from about 20 min, leading to a suspicion of functional gallbladder and Oddi's sphincter disorder. We, therefore, administered a calcium channel blocker to loosen Oddi's sphincter, but the patient' symptoms did not improve. Endoscopic sphincterotomy (EST) was then performed to abolish Oddi's sphincter, but this also failed to improve her symptoms (Fig. 4a, b). Biliary scintigraphy was repeated to confirm bile flow and, as in the first case, radionuclide accumulation increased over time (Fig. 4c).

Based on these results, we diagnosed dyskinesia of the gallbladder and recommended cholecystectomy; however, the patient refused to undergo cholecystectomy immediately because she was uncertain if the treatment would be effective after such a long history of disease. The patient had a strongly flexed cystic duct, which is thought to be the cause of gallbladder dyskinesia, and EGBS was performed to try to correct the flexed part of the cystic duct (Fig. 5a, b). After EGBS, patient's symptoms improved remarkably. A third biliary scintigraphy was performed to determine the efficacy of the treatment, and showed that radionuclides in the gallbladder and common bile duct accumulate up to 30 min, but after 30 min, the radionuclides in the gallbladder decrease due to excretion into the duodenum (Fig. 5c). This indicated that cholecystectomy would be effective, and cholecystectomy was accordingly performed on a standby basis. The patient's subsequent course was good, and a laparoscopic cholecystectomy was performed two months after EGBS. After laparoscopic cholecystectomy, there was no recurrence of symptoms such as right quadrant pain and vomiting up to 8 months, and the patient continues to be followed clinically.

Discussion

Gallbladder dyskinesia is a disease in which functional abnormalities of bile flow cause gallstone attack-like symptoms, such as abdominal pain and vomiting, in the absence of any organic abnormality in the biliary system, including the gallbladder. It is classified as a functional gallbladder disorder according to the Functional Gallbladder and Sphincter of Oddi Functional Gastrointestinal Disorders in the Rome IV diagnostic criteria [1]. Bile produced in the liver is secreted into the intrahepatic bile ducts, concentrated in the gallbladder, and excreted via the papilla into

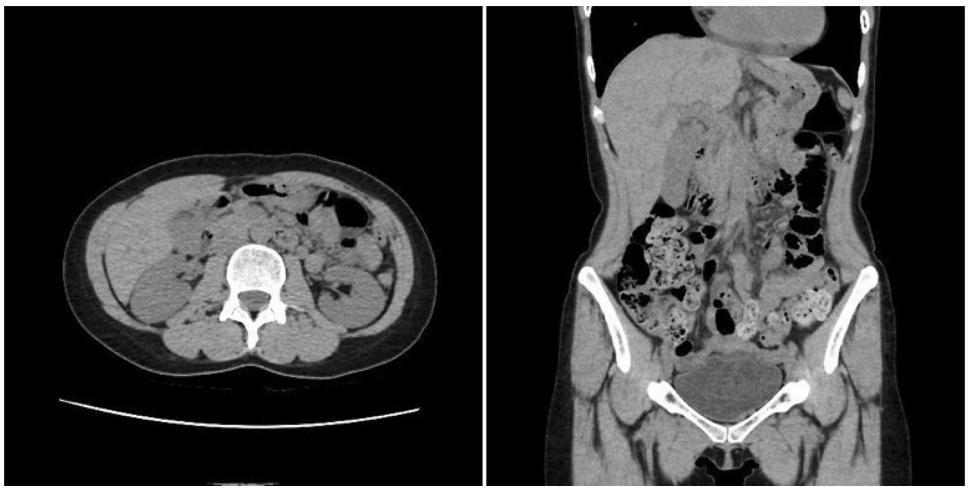

Fig. 1 Abdominal plain computed tomography at first medical examination. There were no stones or strictures in the biliary tract.

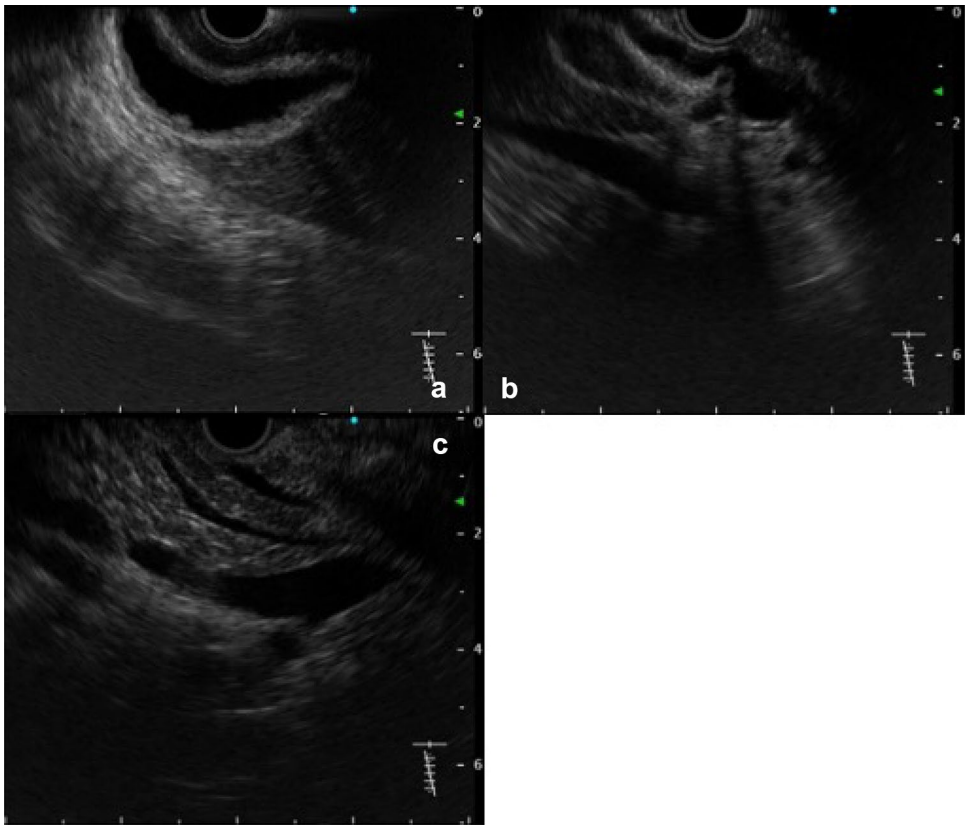

Fig. 2 Endoscopic ultrasound images after first medical examination are shown. **a** An image of the gallbladder observed from the duodenal bulb. Endoscopic ultrasound showed no gallstones and debris in the gallbladder. The wall of the gallbladder was thickened and a 2 mm polyp was observed. **b** An image of the bifurcation of the cystic duct observed from the duodenal bulb. There was no obvious choledochal stone or stenosis in the common bile duct and cystic duct. **c** An image of the common bile duct and main pancreatic duct observed from the descending part of the duodenum. Pancreaticobiliary maljunction was not observed, and no other abnormalities including choledochal stone and stenosis were found

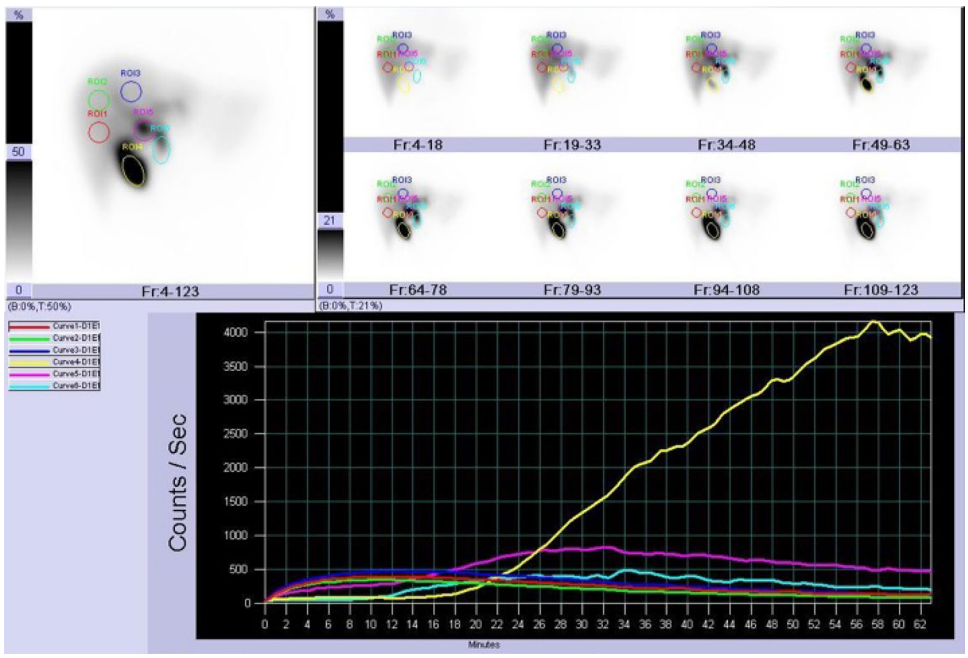

Fig. 3 In the first biliary scintigraphy, the figure shows the amount of radionuclides over time. It can be seen that the radionuclides accumulate in the gallbladder (yellow) without decreasing over time. Radionuclides in the cystic duct (purple), common bile duct (light blue), and intrahepatic bile duct (red, green and blue) do not accumulate, and there is no ejection into the duodenum

the duodenum. Cholecystokinin, a gastrointestinal hormone secreted from the duodenum, is involved in the coordinated movement of the gallbladder, Oddi's sphincter, and duodenum at this time, causing the gallbladder to contract and Oddi's sphincter to relax. This process is believed to be strongly involved in bile-excretion disorders [2].

The Rome IV classification is based on the diagnostic criteria of biliary pain and absence of gallstones or other

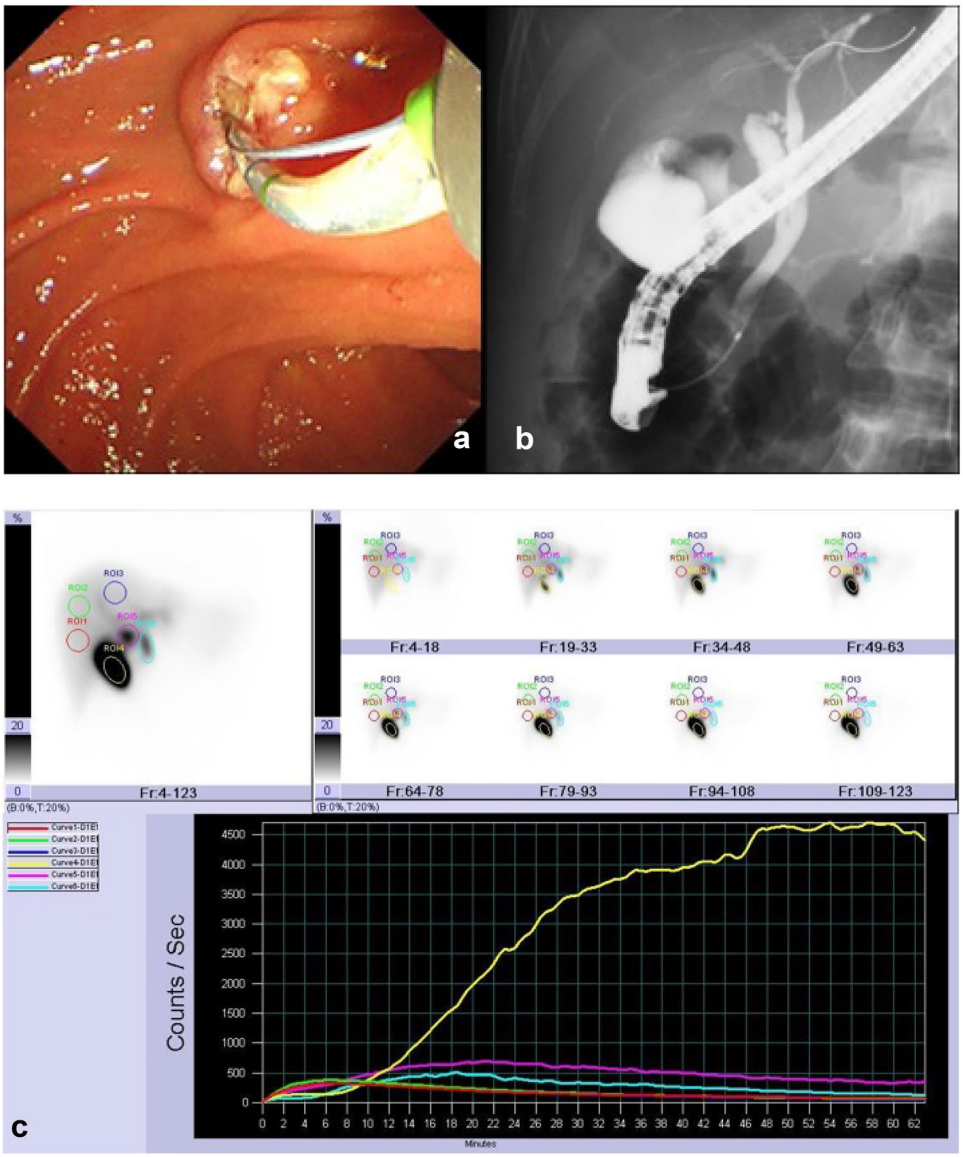

Fig. 4 The images at the time of endoscopy when EST was performed and biliary scintigraphy after treatment are shown. **a** EST was performed to abolish Oddi's sphincter. A small incision was made in this case. **b** Fluoroscopic images during the first endoscopic retrograde cholangiopancreatography. Flexion of the cystic duct was observed. **c** Biliary scintigraphy after EST also shows that radionuclides accumulate in the gallbladder (yellow) without decreasing with time. Radionuclides in the cystic duct (purple), common bile duct (light blue), and intrahepatic bile duct (red, green and blue) do not accumulate and there is no ejection into the duodenum

organic disease, with low biliary ejection fraction on biliary scintigraphy and normal blood enzymes, bilirubin, and amylase/lipase as supplementary diagnoses. The diagnosis of gallbladder dyskinesia is suggested when the above criteria are fulfilled [1].

Biliary scintigraphy using 99 m technetium is commonly used to evaluate the gallbladder ejection fraction. Given that changes in radioisotopes over time reflect bile flow, changes can, thus, be expressed semi-quantitatively by the number of γ-rays [3–5]. However, other diseases, including functional gastrointestinal disorders, can also cause delayed excretion, and the Rome IV diagnostic criteria emphasize the importance of patient selection in the decision to perform biliary scintigraphy. In addition, although there is no correlation between biliary ejection fraction and Oddi's sphincter pressure, elevated sphincter pressure has been reported in patients with gallbladder dyskinesia, which may make it difficult to differentiate between gallbladder dyskinesia and functional Oddi's sphincter disorder [6]. In the current case, initial biliary scintigraphy showed excretion of radionuclides into the common bile duct after 15 min and EST was, thus, chosen as the treatment; however, there was no improvement in the patient's symptoms. A diagnosis of gallbladder dyskinesia was therefore made based on residual delayed bile excretion from the gallbladder on biliary scintigraphy, performed to confirm the efficacy of treatment.

Treatment of gallbladder dyskinesia involves pharmacotherapy with anticonvulsants and neuromodulators, but cholecystectomy is considered if these measures are ineffective [7–9]. Although cholecystectomy has been reported to be effective in more than 80% of patients with gallbladder

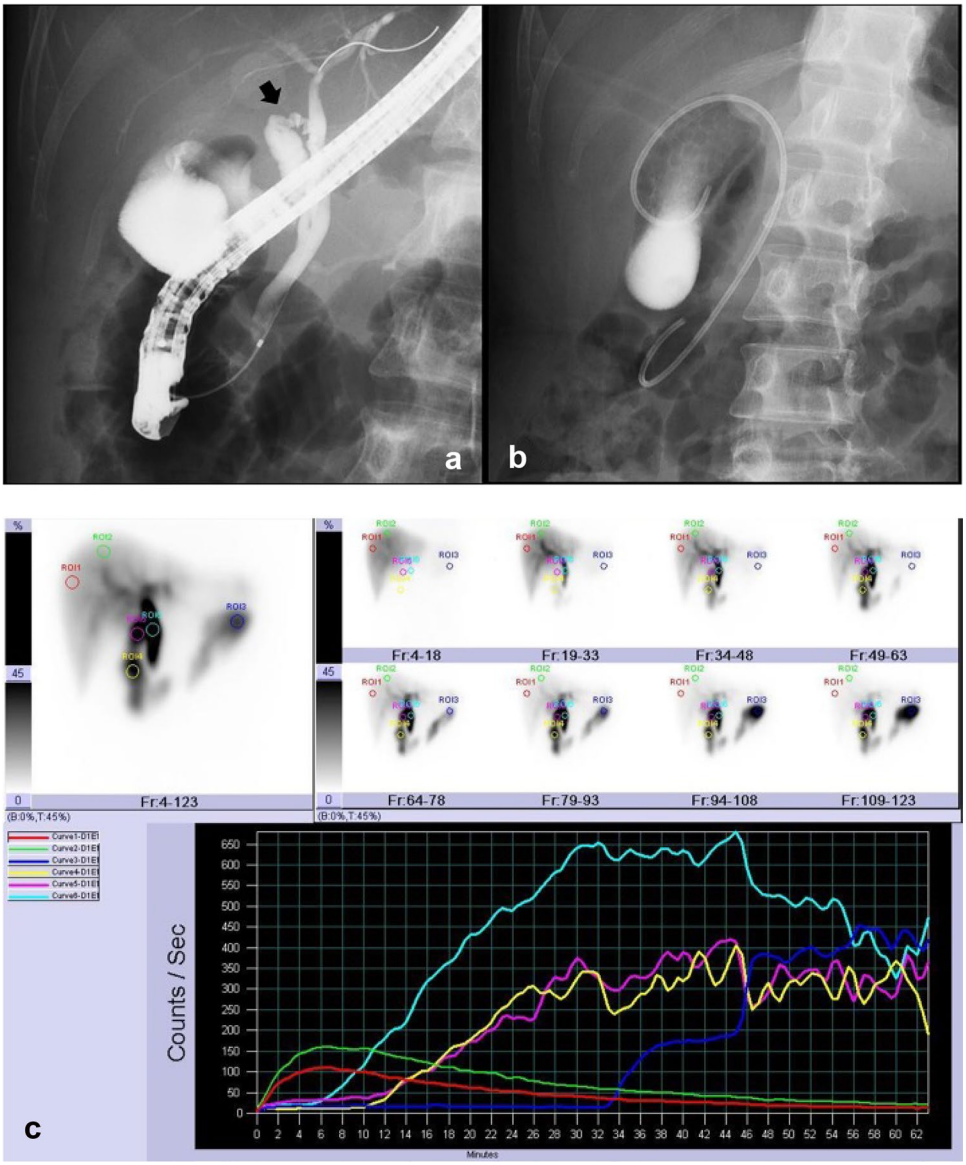

Fig. 5 Images at the time of endoscopy when EGBS was performed and biliary scintigraphy after treatment are shown. **a** Endoscopic retrograde cholangiopancreatography images shows the strongly flexed cystic duct (arrow). **b** Endoscopic retrograde cholangiopancreatography image when EGBS was performed. A Zimmon (Cook Group Inc., IN, USA) 7 Fr, 14 cm plastic stent was placed into the gallbladder. **c** Biliary scintigraphy after EGBS shows that radionuclides flow into the gallbladder (yellow) for up to 20 min. In contrast, there is no accumulation of radionuclides in the intrahepatic bile duct (red, green) or gallbladder, and radionuclides are ejected into the cystic duct (purple), common bile duct (light blue) and duodenum (blue)

dyskinesia, there has only been one report of a randomized trial, and further investigations are therefore needed [7, 10].

In the present case, cholecystectomy was recommended after the diagnosis of gallbladder dyskinesia but the patient was hesitant to undergo surgery, and EGBS was, therefore, performed to improve gallbladder bile outflow. This helped to confirm the diagnosis of gallbladder dyskinesia and predicted the efficacy of cholecystectomy.

Conclusion

In conclusion, this is the first report of a patient with gallbladder dyskinesia in whom treatment response was confirmed by biliary scintigraphy before and after EGBS. This case suggests that that EGBS may be an effective procedure for supporting the diagnosis and treatment of gallbladder dyskinesia.

Acknowledgements We thank Susan Furness, PhD, from Edanz (https://jp.edanz.com/ac) for editing a draft of this manuscript.

Funding None.

Declarations

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Cotton PB, Elta GH, Carter CR, et al. Rome IV. Gallbladder and sphincter of Oddi disorders. Gastroenterology. 2016;1150:1420–9. https://doi.org/10.1053/j.gastro.2016.02.033. PMID: 27144629
2. Toouli J. Biliary dyskinesia. Curr Treat Options Gastroenterol. 2002;5:285–91.
3. Greenberg JJ. What is biliary hyperkinesia? Glob Surg. 2018. https://doi.org/10.15761/GOS.1000174.
4. Harvey RF, Read AE. Effect of cholecystokinin on colonic motility and symptoms in patients with the irritable-bowel syndrome. Lancet. 1973;1:1–3.
5. Fink-Bennett D, DeRidder P, Kolozsi W, et al. Cholecystokinin cholescintigraphic findings in the cystic duct syndrome. J Nucl Med. 1985;26:1123–8.
6. Ruffolo TA, Sherman S, Lehman GA, et al. Gallbladder ejection fraction and its relationship to sphincter of Oddi dysfunction. Dig Dis Sci. 1994;39:289–92.
7. Bielefeldt K, Saligram S, Zickmund SL, et al. Cholecystectomy for biliary dyskinesia: how did we get there? Dig Dis Sci. 2014;59:2850–63.
8. Bar-Meir S, Halpern Z, Bardan E. Nitrate therapy in a patient with papillary dysfunction. Am J Gastroenterol. 1983;78:94–5.
9. Guelrud M, Mendoza S, Rossiter G, et al. Effect of nifedipine on sphincter of Oddi motor activity: studies in healthy volunteers and patients with biliary dyskinesia. Gastroenterology. 1988;95:1050–5.
10. Veenstra BR, Deal RA, Redondo RE, et al. Long-term efficacy of laparoscopic cholecystectomy for the treatment of biliary dyskinesia. Am J Surg. 2014;207:366–70 (**discussion 369–70**).