

Cystic duct cyst: Report of one case

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Cystic duct cyst is an extremely rare anomaly. The classification developed by Tadoni is used for the other bile duct anomalies, but cystic duct cysts are excluded from this classification. In this case report, acute pancreatitis was diagnosed based on the clinical and laboratory findings of the patient, who presented with a complaint of nonspecific abdominal pain. Cystic duct cyst was determined with magnetic resonance cholangiopancreatography performed after determination of a cystic lesion on the neck of the gallbladder by abdominal ultrasonography. Cystic duct cyst is an extremely rare anomaly, and advanced examination is required for the diagnosis.

Key words: Cystic duct, cyst, congenital

Sistik kanal kisti; bir olgu sunumu

Sistik kanal kisti oldukça nadir görülen bir anomalidir. Diğer safra kanal anomalilerinin sınıflandırılmasında, Tadoni tarafından yapılan sınıflama kullanılırken, sistik kanal kistleri bu sınıflamanın dışında tutulmaktadır. Bu olgu sunumunda nonspesifik karın ağrısı şikayeti ile başvuran hastada klinik ve laboratuvar bulguları ile akut pankreatit tanısı konuldu. Hastanın yapılan batin ultrasonografisinde safra kesesi boyun kısmında kistik lezyon tespit edilmesi üzerine yapılan manyetik rezonans kolanjiopankreatografi'de sistik kanal kisti tespit edildi. Sistik kanal kisti oldukça nadir görülen bir anomali olup ve tanı için ileri tetkik gerekmektedir.

Anahtar kelimeler: Sistik kanal, kist, konjenital

INTRODUCTION

Bile duct cysts are rare congenital anomalies, characterized by cystic dilatation of intrahepatic and/or extrahepatic bile ducts (1). Bile duct cysts were classified by Alonso-Lej et al. (2) and modified by Todani et al. (3). However, this classification does not include cystic duct dilatations. Cystic duct cyst (CDC) is rarer than the other bile duct cysts (4). In this case report, a patient with an extremely rare CDC is discussed.

CASE REPORT

In this case report, a 47-year-old male applied to the Emergency Department with complaints of waistband type abdominal pain and nausea,

which had started the day before. The physical examination of the patient revealed arterial blood pressure: 130/80 mmHg and pulse: 82/min. Respiratory sounds were normal on auscultation, and sensitivity was determined during epigastric and right upper quadrant abdominal examinations. Murphy finding was negative. Total thyroidectomy due to multinodular goiter one year before and 100 µg levothyroxine-sodium administrations once per day in the postoperative period were notable in the medical history. Laboratory examinations determined: white blood cells (WBC) 6110/mm³, hemoglobin (Hb) 14.4 g/dl, hematocrit (Hct) 43.5%, glucose 92.1 mg/dl, amylase 2962, as-

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Manuscript received: 03.02.2012 **Accepted:** 08.04.2012

Turk J Gastroenterol 2013; 24 (2): 170-172
 doi: 10.4318/tjg.2013.0521

partate aminotransferase (AST) 685.8 mU/ml, alanine aminotransferase (ALT) 1054.1 mU/ml, total/direct bilirubin 3.147/1.79 mg/ml, lactate dehydrogenase (LDH) 549 mU/ml, and C-reactive protein (CRP) 0.905. Wall thickness of the gallbladder was determined to be 3 mm, and a dense cystic lesion measuring 31x16 mm in the area of the calculus – the largest 6 mm – and neck was determined in the abdominal ultrasonography (USG). The patient was hospitalized with a diagnosis of Ranson 2 biliary pancreatitis based on the clinical and laboratory findings. Hydration, pantoprazole, metoclopramide, and diclofenac sodium treatment were applied the first day after oral intake was stopped because the patient complained of nausea. Since the patient's symptoms improved during the follow-up, treatment was continued after oral food intake was started on the first day. Magnetic resonance cholangiopancreatography (MRCP) was performed. A cystic lesion (CDC) measuring 41x22 mm was monitored with MRCP. MRCP image of the patient is shown in Figure 1.

Upon improvement in the patient's clinical and laboratory findings, he was discharged from the hospital on the 5th day of hospitalization. Elective surgery due to symptomatic cholelithiasis and CDC was suggested to the patient.

DISCUSSION

Choledochal cysts are rare congenital anomalies of the biliary tree (4-6). Incidence at birth ranges

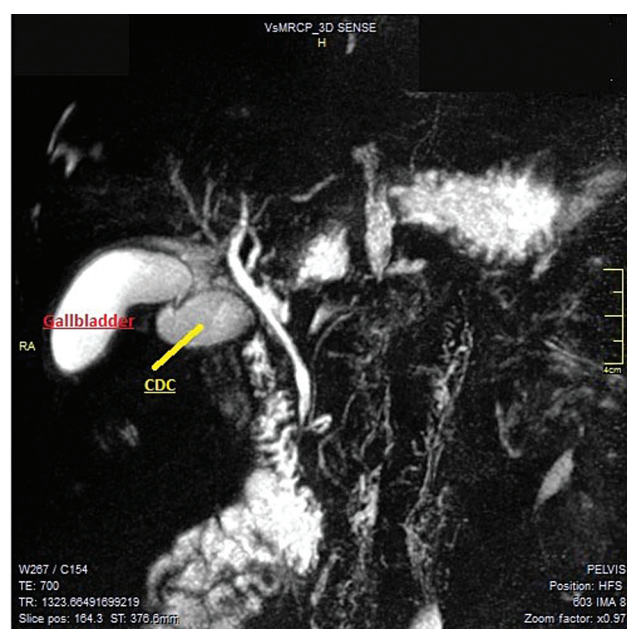


Figure 1. MRCP image of the patient.

between 1/100000 and 1/150000 in the West (4,6,7). While the incidence rate in the United States of America and Australia is 1/13500 and 1/15000, respectively, the incidence is extremely high in Asia, especially in Japan (1/1000). However, the reason for the high incidence in Asia is still not known. It is more frequently seen in females (7). The diagnosis is usually established in newborns and in childhood. The diagnosis is established in adulthood in 20-30% of the cases (4,6,7).

Bile duct cysts were defined for the first time by Alonso-Lej et al. in 1959 (2). This classification was modified by Tadoni et al. (3) in 1977. This classification is widely used in clinics today. Bile duct cysts are divided into five major groups as Types I-V. Type I is divided into three subgroups as Types IA, IB, IC, and Type IV is divided into two subgroups as Types IVA and IVB. Type V is also defined as Caroli disease. The most frequent types are, respectively, Type I and Type IV (7). CDCs are rarer than bile duct cysts and are excluded from this classification. In the case report presented by Baj et al. (4) in 2002, they indicated that there were eight CDCs reported in the literature. Chan et al. (8), in their 2009 study, indicated that there were 14 CDCs in the literature.

Clinical symptoms are nonspecific. Asymptomatic or right upper quadrant pain may be observed in adults (6,9). Surgery is performed in approximately 50% of the patients before diagnosis. While icterus is rarely seen in adults, cholecystitis, cholangitis and pancreatitis are observed more frequently. A palpable abdominal mass is seen rarely (6,10). Diagnosis of bile duct anomalies is difficult and radiological examination is required. USG, MRCP, and endoscopic retrograde cholangiopancreatography (ERCP) are the frequently used examinations for this purpose (3, 6). ERCP is one of the methods that may be chosen for both diagnosis and treatment (11). The patient in this case report presented with nonspecific right upper quadrant pain, and pancreatitis was determined based on the laboratory examinations. MRCP was planned after determination of a cystic lesion on the neck of the gallbladder along with gallbladder calculus in USG, and the diagnosis was confirmed with MRCP.

The cancer risk in bile duct cysts is stated to increase with age (6,12). Therefore, surgical resection is suggested in the treatment of bile duct cysts (4,6).

Baj et al. (4) in their case report presented a 50-year-old female who presented with the complaint of right upper quadrant pain that continued for six years; the anamnesis included thyroxine supplementation for autoimmune thyroiditis. CDC was diagnosed with cholecystographic helical computed tomography after cystic lesion determination between the gallbladder and common hepatic duct with USG. However, the patient refused surgical treatment. Our patient's complaints at presentation were different from those of Baj et al.'s case. When the cystic lesion was detected on USG, CDC diagnosis was established with MRCP. Chan et al. (8) reported that laparoscopic cholecystectomy

was planned after Type II choledochal cyst was diagnosed by MRCP during the routine follow-up of a 41-year-old male with a history of nephrolithiasis and cholelithiasis. However, CDC, not Type II choledochal cyst, was determined by laparoscopic cholecystectomy. The patient mentioned in this case report was the 15th patient with CDC reported in the literature, and laparoscopic cholecystectomy was stated to be a safe and effective method in patients with CDC. In this case report, CDC was categorized as Type VI.

In conclusion, CDC is an extremely rare anomaly, and advanced examination is required for the diagnosis.

REFERENCES

- Mabrut JY, Bozio G, Hubert C, Gigot JF. Management of congenital bile duct cysts. *Dig Surg* 2010; 27: 12-8.
- Alonso-Lej F, Rewer WB Jr, Pessagno DJ. Congenital choledochal cyst, with a report of 2, and an analysis of 94, cases. *Int Abstr Surg* 1958;108:1-30.
- Todani T, Watanabe Y, Toki A, et al. Co-existing biliary anomalies and anatomical variants in choledochal cyst. *Br J Surg* 1998; 85: 760-3.
- Baj M, Sengupta S, O'Donnell L. Choledochoceles of the cystic duct: a case report. *Internet Journal of Gastroenterology* 2002; 1.
- Tian Y, Wu SD, Zhu AD, Chen DX. Management of type I choledochal cyst in adult: totally laparoscopic resection and Roux-en-Y hepaticoenterostomy. *J Gastrointest Surg* 2010; 14: 1381-8.
- Geraci G, Nigro CL, Sciuto A, et al. Surgical treatment of choledochal cyst associated with an aberrant posterior hepatic duct: report of a case and brief literature review. *Case Rep Gastroenterol* 2011; 5: 73-81.
- Singham J, Yoshida EM, Scudamore CH. Choledochal cysts: part 1 of 3: classification and pathogenesis. *Can J Surg* 2009; 52: 434-40.
- Chan ES, Auyang ED, Hungness ES. Laparoscopic management of a cystic duct cyst. *JLS* 2009; 13: 436-40.
- Jordan PH Jr, Goss JA Jr, Rosenberg WR, Woods KL. Some consideration for management of choledochal cysts. *Am J Surg* 2004; 187: 790-5.
- Söreide K, Körner H, Havnen J, Söreide JA. Bile duct cysts in adult. *Br J Surg* 2004; 91: 1538-48.
- Garrido A, León R, López J, Márquez JL. [An exceptional case of choledochoceles and pancreas divisum in an elderly man]. *Gastroenterol Hepatol* 2012; 35: 8-11.
- Okada A, Hasegawa T, Oguchi Y, et al. Recent advances in pathophysiology and surgical treatment of congenital dilatation of the bile duct. *J Hepatobiliary Pancreat Surg* 2002; 9: 342-51.