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Case report
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A rare case: Asymptomatic spontaneous pneumobilia

Nadir bir olgu: Asemptomatik spontan pnömobilya

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Abstract

Pneumobilia, or aerobilia, is defined as the presence of gas in the biliary system. It may occur for various reasons, including recent enterobiliary system interventions, sphincter of oddi dysfunction, and enterobiliary fistula and anastomoses. A 61-year-old male patient presented to our emergency department due to nausea and vomiting. He had no history of drug use other than proton pump inhibitor medication, nor of alcohol or substance use. Although no history of previous medical intervention was present, gas (pneumobilia) was detected in the intrahepatic biliary ducts. We suspected that the incidentally detected pneumobilia in this patient, who also suffered from food poisoning, developed spontaneously in association with peptic ulcer and might have derived from a thin, asymptomatic enterobiliary fistula. Depending on its cause, pneumobilia may not require emergency surgery. The possibility of enterobiliary fistula should not be overlooked in patients presenting to the emergency department with chronic peptic ulcer symptoms.

Keywords: Pneumobilia, Gas in intrahepatic bile ducts, Saber sign, Hepatic portal vein gas

Öz

Aerobilia olarak da bilinen pneumobilia, biliyer sistemde gaz olması olarak tanımlanır. Son zamanlarda yapılmış enterobilier sistem girişimleri, Oddi sfinkter disfonksiyonu, enterobilier sistem fistül ve anastomozları gibi değişik nedenlerle gelişebilir. Altmış bir yaşında erkek hasta bulantı kusma şikayeti ile acil servise başvurdu. Proton pompa inhibitörü ilacı dışında herhangi bir ilaç, alkol veya madde kullanım öyküsü olmayan hastada, daha önce herhangi tıbbi girişim işlemi geçirmemiş olmamasına rağmen intrahepatik safra yollarında gaz (pneumobilia) tespit edildi. Besin zehirlenmesi de olan hastada tesadüfen tespit edilen pneumobilia'nın, peptik ülsere bağlı spontan gelişmiş, asemptomatik enterobilier ince bir fistülden kaynaklanmış olabileceği düşünüldü. Pneumobilia, geliştiği nedene bağlı olarak acil cerrahi girişim gerektirmeyebilir. Kronik peptik ülser şikayetleri ile acil servise müracaat eden hastalarda enterobilier fistül gelişmiş olabileceği gözardı edilmemelidir.

Anahtar kelimeler: Pneumobilia, İntrahepatik safra yollarında gaz, Saber işareti, Hepatik portal ven gazı

Introduction

Pneumobilia, or aerobilia, is defined as the presence of gas in the biliary system. The condition has a broad etiological spectrum, including recent enterobiliary system interventions, drugs and structural causes leading to sphincter of Oddi dysfunction, spontaneous and surgical enterobiliary fistulae, malignancy, and rarely infections and biliary-bronchopleural fistula [1]. We report a case presenting to the emergency department with nausea and vomiting and diagnosed with rare spontaneous pneumobilia, together with the current literature.

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Case presentation

A 61-year-old male patient, from whom verbal consent for this case report was obtained, presented to our emergency department due to nausea and vomiting. On arrival, his general condition was average, and he was conscious and cooperative. Arterial blood pressure was 100/60mmHg, heart rate 88/min, and body temperature 36.7°C, and respiration was normal. Peptic ulcer was present in his medical history, and he had been using proton pump inhibitors (PPIs) irregularly for 7-8 years. He had no history of alcohol or substance use, nor of previous medical intervention. The patient reported occasional recurrence of epigastric pain, with partial improvement after taking PPIs, that nausea and vomiting began a few hours after eating in the evening, and that no diarrhea occurred. No positive signs other than epigastric tenderness not radiating to the back were present at physical examination. Normal sinus rhythm was present at echocardiography (EKG). No pathological finding was determined at lung x-ray. Symptomatic intravenous (IV) 10 mg metoclopramide and 40 mg pantoprazole were administered with fluid infusion. At blood tests, glucose was 165 mg/dl (range 74-106 mg/dl), amylase 164 U/L (range 28-100U/L), and gamma glutamyl transferase (GGT) 71 U/L (<55). Hepatic enzymes, bilirubin values, C-reactive protein and other biochemical parameters were within normal reference ranges. Leukocyte value at complete blood count was 12.770/mm3 (range 3980-10.200/mm3). Blood gas oxygen saturation was 95.9%, carbon dioxide pressure was 43.1 mmHg, and pH was 7.49. Abdominal ultrasound (USG) revealed gas in the intrahepatic bile ducts (pneumobilia). The gall bladder and choledochal duct wall were normal, and no dilation was determined (Figure 1). Air was observed in the intrahepatic bile ducts at both non-contrast and oral and iv with-contrast computerized tomography (CT), but no stone, mass, fistula, perforation or stenosis were observed in the hepatobiliary and enterobiliary tract. No appearance of air or fluid was encountered in the abdomen (Figure 2). We suspected that the patient's acute onset symptoms were due to food poisoning, but that the pneumobilia observed might derive from the asymptomatic, fine enterobiliary fistula developing in association with peptic ulcer present in his previous history. The general surgery department was consulted, but no condition requiring emergency surgery was determined. The peptic ulcer again improved following symptomatic treatment. The patient was discharged, and polyclinic check-up visits were advised for advanced tests and treatment.

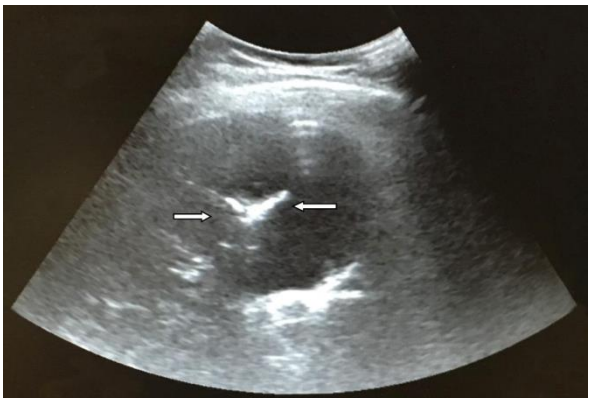

Figure 1: The image of the pneumobilia on the ultrasound of the patient

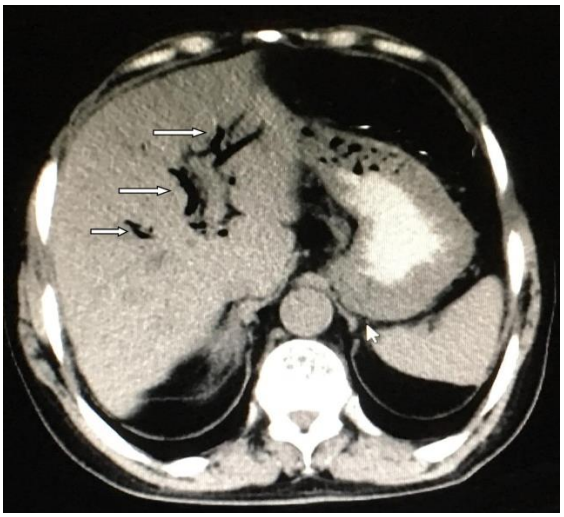

Figure 2: The image of the pneumobilia on the CT of the patient

Discussion

Pneumobilia is a rare condition. Due to the limited number of studies available, we found no up-to-date data concerning its incidence. Yamashita et al. [2] noted a relation between pneumobilia and the enterobiliary system, and reported the presence of spontaneous bilioenteric fistula in 33 (1.9%) out of 1929 patients with existing biliary disease, and that 15 (44%) of these patients with bilioenteric fistula presented due to pneumobilia.

In their retrospective study, Lassandro et al. [3] determined pneumobilia detection rates of 37.04% at abdominal x-ray, 55.6% at abdominal ultrasound, and 88.89% at abdominal CT. In the present case, the air in the patient's intrahepatic bile ducts could not be detected at x-ray imaging, but was clear at USG and CT.

Radiologically, pneumobilia can be confused with hepatic portal vein gas. The air in pneumobilia is in the direction of the bile flow, and has fewer branches. On x-ray images this is known as the sabre sign, and is a reliable indication at differential diagnosis. In contrast, portal venous gas is retrograde to the bile flow, extending both to the hepatic hilum region and toward Glisson's capsule, and exhibits a tree-like appearance in the peripheral branches [4,5]. Hepatic artery calcification can also mimic pneumobilia [6].

Our scan of the literature various case reports of pneumobilia development. It has been reported in association with emphysematous cholecystitis, emphysematous pyelonephritis, hepatic abscess, and intestinal tuberculosis [1,7,8]. Some case reports have shown that it can develop in association with metastatic colon cancer, as well as lymphoma, and may be associated with malignancy [9,10]. Gallstone is one important cause of pneumobilia, representing one in four bowel obstructions, and pneumobilia has been detected in 89% of these ileus cases [3]. Gallstone can also lead to pneumobilia without causing ileus [1]. One report described pneumobilia caused by choledochogastric fistula developing in association with congenital gall bladder agenesis [11]. Pneumobilia can also develop as a complication of invasive diagnostic procedures, such as endoscopic retrograde cholangiopancreatography (ERCP), or following surgical procedures [1]. It may develop in association with blunt and penetrating traumas, and there is a case report of cardiopulmonary resuscitation-related

development. This was thought to derive from the retrograde passage of air to the biliary system due to increased intra-abdominal pressure associated with blunt trauma [12-14].

Incidentally-detected pneumobilia can be assessed as a guide to diagnosis. In one case, enterobiliary fistula was suspected due to pneumobilia detected incidentally at USG in a case presenting due to diarrhea, and cholecystocolic fistula was determined with endoscopic cholangiopancreatography [15]. In another case, pneumobilia was observed incidentally in a patient presenting due to peptic ulcer symptoms persisting despite long-term medication. Cholecystoduodenal fistula was detected at endoscopy performed in order to identify the source of the pneumobilia [16]. In the present case, acute onset nausea and vomiting symptoms were attributed to food poisoning. However, the history of chronic peptic ulcer and irregular PPI use suggested that the enterobiliary fistula may have developed in association with peptic ulcer. No enterobiliary fistula was observed at multislice spiral computed tomography performed with both oral and iv contrast material.

Both diagnosis and treatment can be performed with endoscopic procedures in some patients with pneumobilia thought to be associated with enterobiliary fistula that cannot be detected at CT. The defective region can be closed by inserting an endobiliary prosthesis with ERCP. Magnetic resonance cholangiopancreatography (MRCP) can show the site of the bile leak and fistula in detail in a non-invasive manner [17,18]. No endoscopic or magnetic resonance imaging was performed in the emergency department in the present case. Some gastrobiliary fistulas identified endoscopically have been reported to close spontaneously with anti-ulcer treatment [19]. Emergency surgery was not considered. The patient was discharged, and polyclinic check-up visits were advised for advanced tests and treatment.

Conclusion

The etiological cause can be evaluated by establishing a relation between incidentally detected pneumobilia and detailed history and clinical findings, and the decision whether emergency surgical intervention is required can be made accordingly. The possibility of enterobiliary fistula should also be considered in patients presenting to the emergency department with symptom of chronic peptic ulcer, and these should be referred to the polyclinic for advanced tests.

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