

## Acute abdominal disease associated with gallbladder torsion recovered after cholecystectomy: a rare case report and literature review

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**Background:** Gallbladder torsion is very rare and easily misdiagnosed as biliary disease. It is defined as the rotation of the gallbladder along the axis of the cystic pedicle on the mesentery. As gallbladder rotation involves the gallbladder artery, the blood supply is blocked, resulting in gallbladder ischemia and eventual necrosis. If misdiagnosis occurs and treatment is delayed, gallbladder torsion can develop into a lethal disease. The typical imaging features of gallbladder torsion in this case are a good learning resource for our young physicians, as well as providing a rare, unusual and typical case for our current literature database.

**Case Description:** We present a rare case of gallbladder torsion in a 19-year-old man. The patient complained of sudden recurrent pain and discomfort in the right upper abdomen with vomiting for 12 hours. Abdominal ultrasound and computed tomography (CT) scan showed gallbladder enlargement and signs of acute cholecystitis in emergency examination, and there were no signs of cholecystolithiasis. Considering that the patient was a young male and the patients prefer conservative treatment, symptomatic treatment was given. However, there was no obvious effect after 1 day of medical treatment, but severe abdominal pain in the upper right quadrant continues to progress. Finally, the patient underwent laparoscopic cholecystectomy, and the gallbladder was found to be enlarged with ischemic necrosis, which was caused by gallbladder torsion. The patient recovered 2 days after surgery and was discharged without complications.

**Conclusions:** Although the clinical manifestation is similar to that of typical acute calculous cholecystitis, gallbladder torsion can be diagnosed early through some special signs on imaging examination, such as distorted cystic duct signs ("beak and whirl" sign), gallbladder dilatation with gallbladder fossa effusion, and gallbladder in the horizontal position. These signs can help primary surgical treatment and prevent fatal complications such as gallbladder gangrene, perforation, and biliary peritonitis. Therefore, for inexperienced doctors, careful imaging features are required for the correct diagnosis of rare gallbladder torsion.

Keywords: Gallbladder torsion; acute abdominal disease; cholecystitis; case report

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#### Introduction

Depending on the related literature, it is reported that the incidence of gallbladder torsion is about 1 in 350,000 hospitalized patients (1). Generally speaking, gallbladder torsion is more common in adult patients (84% over 18 years old), and the proportion of women is higher than that of men (2). As the gall bladder rotates, the cystic artery becomes involved, blocking the blood supply. Early diagnosis is important since this rare morbid structure can develop rapidly, leading to sepsis and septic shock, and the mortality rate can reach 6% (3). In this case report, the preoperative diagnosis of gallbladder torsion was not clear. Of the more than 500 case reports published worldwide, only 10% of patients were diagnosed before surgery (3,4). Although there are difficult in preoperative diagnosis, we can still obtain diagnostic clues from typical ultrasound and abdominal computed tomography (CT). We hope that by describing this case we can contribute to the accumulation of a typical image database of this rare lesion, and help inexperienced clinicians to perform preoperative evaluation of this uncommon condition, so as to obtain a complete and accurate diagnosis. We present the following case in accordance with the CARE reporting checklist (available at https://atm.amegroups.com/article/view/10.21037/atm-22-1425/rc).

#### **Case presentation**

A 19-year-old man was admitted to the emergency department for 12 hours with recurrent right upper quadrant abdominal pain in March 2021. The pain was severe, persistent, and confined to the right abdomen. The patient experienced vomiting and mild fever (37.8 °C). Past medical, previous surgical history, drug and family histories were not significant. In the physical examination, all results of vital indexes were steady, and there was no abdominal mass. The patient had right upper quadrant tenderness, mild rebound pain, and was Murphy sign positive, with no other significant tenderness and rebound pain upon abdominal physical examination. The timeline depicts the key information of the patient's history in chronological sequence (Figure 1A). Emergency abdominal ultrasonography showed that the internal diameter of the gallbladder was 75×35 mm, the thickness of the gallbladder wall was 8.3 mm, and the echo of the cystic wall showed a multi-layered weak echo zone, in which densely dotted echo could be seen, and there was no dark area of effusion

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around the gallbladder. Importantly, we did not see signs of gallstones (Figure 1B). The hemogram hinted that the patient's white blood cells were 19.4×10<sup>9</sup>/L, neutrophils 82.6%, and c-reactive protein 167 mg/L. Other blood biochemical indexes, including liver enzymes and pancreatic enzymes, had no abnormal changes. Taking into account these findings, we considered the diagnosis of acute cholecystitis without calculus and abnormal enlargement of the gallbladder. Considering that the patient was a young male and that patients prefer conservative treatment, empirical antibiotic treatment was given based on intravenous drip of 1.0 g Latamoxef Sodium for injection, twice per day. Other symptomatic treatments include pain relief, antiemetic therapy, correction of electrolyte disorders and resuscitation therapy. After 1 day of antibiotics and symptomatic treatment, the right upper quadrant pain was not significantly relieved, with signs of continued worsening pain. The patient then underwent an abdominal CT scan, which revealed a sudden narrowing of the complex structure between the gallbladder and the cystic duct, similar to a "beak". At the same time, the gallbladder was enlarged. The wall of the gallbladder was thickened, and there was no abnormal density in the gallbladder cavity (Figure 1C-1E).

Based on these unusual imaging findings, we recommended laparoscopic exploratory surgery. Under laparoscopy, ischemia, blackening, necrosis, and significant enlargement of the gallbladder were observed. In addition, an abnormally enlarged gallbladder measuring approximately  $80 \times 50 \times 40$  mm was seen. Torsion of the gallbladder could be seen after rotating 180° counterclockwise along the axis of the gallbladder vascular pedicle. The gallbladder was suspended to the liver bed only by a thin patch of the mesentery, which causes the gallbladder to move freely (*Figure 2*).

Gallbladder torsion was diagnosed, and cholecystectomy was performed. There was no major intraoperative complications during the operation. Inflammatory indicators on the first postoperative day suggested that the white blood cell count dropped to  $8.49 \times 10^{\circ}$ /L. The patient recovered well without any postoperative complications and was discharged on the second day after surgery. Postoperative pathological results showed full-thickness hemorrhage and necrosis of the gallbladder wall and unclear structure. It was diagnosed as gallbladder ischemic infarction (*Figure 3*). All procedures performed in this study were in accordance with the ethical standards of the institutional and national research committee(s) and with the Declaration of Helsinki (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and

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**Figure 1** The timeline and typical imaging of the case. (A) The timeline of the essential information in the patient's history in chronological sequence. (B) Abdominal ultrasonography. The inner diameter of the gallbladder was 75×35 mm, and the thickness of the cystic wall was 8.3 mm. (C-E) Abdominal CT. Abdominal CT showed the typical "beak sign" and "whirl sign" (white arrow), both of which describe vividly the change in angulation of the distorted cystic pedicle. CT, computed tomography.



**Figure 2** Findings of laparoscopic cholecystectomy. (A) Under laparoscopy, gallbladder ischemia, blackening, necrosis, and obvious volume enlargement can be seen ( $80 \text{ mm} \times 50 \text{ mm} \times 40 \text{ mm}$ ). The long axis of the gallbladder is horizontal. (B) The torsion of the gallbladder can be seen after rotating  $180^{\circ}$  counterclockwise along the axis of the gallbladder vascular pedicle (white arrow). (C) Long cystic duct and gallbladder mesangium (white arrow). (D) Gallbladder bed with small area (yellow part), clipped gallbladder artery (green part), and clipped gallbladder duct (blue part).

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**Figure 3** Gross specimens of the gallbladder: postoperative pathological results showed full-thickness hemorrhage and necrosis of the gallbladder wall and unclear structure. It was diagnosed as gallbladder ischemic infarction.

accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

#### Discussion

Gallbladder torsion is a rare acute abdominal condition that usually presents as a sudden onset of severe right upper quadrant abdominal pain and may also present with a presentation similar to acute appendicitis. The lack of typical imaging features and a presentation similar to acute calculous cholecystitis make gallbladder torsion highly susceptible to misdiagnosis in the preoperative diagnosis. Among the cases we reported, the 19-year-old male with gallbladder torsion is even rarer. The clinical presentation was characterized by sudden onset of severe right upper quadrant abdominal pain, vomiting, and high inflammatory markers. The given antibiotic treatment and symptomatic treatment suggested ineffectiveness and the diagnosis of gallbladder torsion was definitively made during the subsequent laparoscopic exploration.

To date, the etiology of gallbladder torsion remains unclear. Variations in physiology and anatomy may cause the gallbladder to "float" freely in its original position, such as an abnormal mesentery with overly long or abnormal fixation of the gallbladder and liver (5). The gallbladder is allowed to hang or "float" because of decreased visceral fat, hepatic atrophy, and increased elasticity of the gallbladder, which is seen as more common in the elderly.

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Severe gastrointestinal peristalsis near the gallbladder and mechanical susceptibility due to kyphosis scoliosis have been considered as predisposing triggers for gallbladder torsion (6). Only a quarter to a half of patients with gallbladder torsion have gallstones, so gallstones are a concomitant condition and not likely to be the underlying etiology (1,5). According to the gross classification, gallbladder torsion can be divided into 2 classifications: one is that the gallbladder and cystic duct are suspended to the liver through the mesentery, while the other is that the cystic duct is connected to the liver alone (7,8). In our case, the classification corresponds to type I, and the gallbladder was attached to only a small part of the liver. This anatomical abnormality forms an unstable gallbladder.

In clinical manifestations, the symptoms of gallbladder torsion usually include persistent biliary colic, which is very close to acute cholecystitis. The degree of gallbladder torsion, which can be subdivided into partial and complete, determines the severity of the pain. Different classifications of gallbladder rotation have been reported, including incomplete rotation ( $\leq 180^\circ$ ) and partial rotation (>180°). It also includes clockwise and counterclockwise rotation (9). Complete torsion is characterized by acute cholecystitis, which can worsen in a short period of time, resulting in gallbladder gangrene, perforation, and fatal consequences. Two triad signs in clinical symptoms and physical examination can increase the diagnosis rate of gallbladder torsion (10). The first triad sign is vomiting, sudden pain, and clinical symptoms of pain in the right upper quadrant. The second triad sign is a palpable mass in the right upper abdomen and a pulse temperature difference in a patient with non-toxic blood symptoms. In the case we reported here, the hemogram suggested elevated leukocyte count, elevated inflammatory biomarkers, and normal liver function tests, but these are not peculiar manifestations of gallbladder torsion (3,11,12). It is worth noting that the initial conservative treatment was not effective, and the pain in the right upper abdomen continued to worsen after treatment with antibiotics, which often suggests that the symptoms of biliary colic are not simply caused by cholecystitis.

Ultrasound and CT of the right upper quadrant are the preferred imaging methods for diagnosis, despite the fact that typical features may not be present in every case. These radiographic images may suggest a floating gallbladder, calculous cholecystitis, and the being gallbladder located outside the normal hepatic fossa. The following 3 signs on CT scans are powerful indications to help diagnose

gallbladder torsion: (I) Gallbladder dilatation with gallbladder fossa effusion; (II) the "beak and whirl" sign; (III) Gallbladder in a horizontal position (13). Results of other auxiliary imaging examinations have also been reported. The magnetic resonance imaging (MRI) features include: T1-weighted imaging of high-intensity areas in the gallbladder wall, indicating hemorrhagic infarction and necrosis. T2-weighted images can show gallbladder wall necrosis (12). Hepatobiliary immoderately acid (HIDA) scans theoretically show the "bulls-eye" feature of gallbladder torsion. This shows that the cystic duct is not optically filled, indicating that it is blocked (5,14). When patients have 2 triad signs in clinical symptoms and physical examination, and patients have a poor response to antibiotics, gallbladder torsion should be regarded as one of the differential diagnoses of acute calculous cholecystitis (11). When the above features are met, typical imaging will be very useful.

In the patient we reported here, one of the limitations was that gallbladder torsion was not clearly diagnosed preoperatively. In addition, the 24-hour conservative treatment purely was inappropriate and extremely risky, as it was ineffective and instead further aggravated the consequences of gallbladder torsion by delaying surgical treatment. Reviewing the imaging data reported in this rare case, we can find typical imaging features of gallbladder torsion, which provides a learning opportunity for our young physicians as well as a rare, unusual and typical case for the current literature base. We can conclude that the following 6 points are consistent with the characteristics of gallbladder torsion: vomiting, sudden pain in the right upper quadrant of the abdomen, mild fever, ineffective treatment with antibiotics, and the typical "beak and whirl" sign.

### Conclusions

Gallbladder torsion is a rare acute abdominal disease, and is often misdiagnosed as acute calculous cholecystitis. For young inexperienced doctors, the combination of 2 triad signs in clinical symptoms and physical examination as well as typical abdominal ultrasound and CT images can improve the preoperative diagnosis rate of gallbladder torsion. Once gallbladder torsion is diagnosed, cholecystectomy must be performed in time in order to avoid gallbladder gangrene, perforation, and severe peritonitis. Laparoscopy cholecystectomy is recommended. Timely diagnosis and prompt surgical treatment guarantee a favourable prognosis of gallbladder torsion.

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#### Footnote

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*Ethical Statement:* The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and national research committee(s) and with the Declaration of Helsinki (as revised in 2013). 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 editorial office of this journal.

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