Case Report

Hemorrhagic cholecystitis afflicted with glanzmann thrombasthenia patient

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Abstract

Hemorrhagic cholecystitis is a rare and potentially life-threatening condition that requires immediate medical attention. If the gallbladder perforates, it can lead to even more severe complications. We present the case of a 27-year-old man with Glanzmann's thrombasthenia, a rare platelets disorder, who visited the emergency department twice before being diagnosed with this condition. Initially, he complained of ear pain, and later, chest pain. Despite a normal ECG and cardiac markers, he was discharged with non-steroidal anti-inflammatory drugs (NSAIDs) for musculoskeletal pain. However, when he returned with abdominal pain, nausea, and vomiting, an ultrasound revealed signs of cholecystitis. An emergency laparoscopic cholecystectomy was performed, which revealed hemorrhagic cholecystitis.

Keywords: Cholecystitis, glanzmann, hemorrhagic, thrombocytopenia

Introduction

Glanzmann thrombasthenia (GT) is an uncommon hereditary bleeding disorder distinguished by deficiencies, either in quantity or quality, within the platelet integrin αIIbβ3.[1] Despite its primarily hemorrhagic nature, GT has been associated with diverse clinical presentations, including unexpected non-bleeding events.

Cholecystitis is commonly identified in Southeastern Asia, with an estimated prevalence rate varying between 5% and 20%.[2] Hemorrhagic cholecystitis, while an infrequent complication of acute cholecystitis, poses challenges in detection due to its symptoms being easily mistaken for more common diagnoses with a reported incidence of 3.5%.[3] This infrequent condition can cause right upper quadrant pain, and its reported mortality rate is between 15% and 20%.[4]

In instances of acute inflammation, the gallbladder's mucosal necrosis may lead to bleeding from small vessels within the wall. Additionally, the occurrence of cystic artery pseudoaneurysms can contribute to hemobilia. Failure to identify hemorrhagic cholecystitis can lead to significant intraperitoneal hemorrhage and, ultimately, death.

In the most extensive documented collection of cases, anticoagulation was linked to 45% of the instances.[5] Other recorded predisposing factors encompass trauma, cirrhosis, vasculitis, malignancy, the administration of NSAIDs, and chronic renal insufficiency.[6]

In this report, we present a rare case of hemorrhagic cholecystitis in a 40-year-old male with a known history of Glanzmann thrombasthenia. This case highlights the necessity for heightened awareness and understanding of rare hematological disorders and

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Received: 20-05-2024 **Revised:** 08-07-2024
Accepted: 15-07-2024 **Published:** 18-11-2024

Access this article online

Quick Response Code:

Website: http://journals.lww.com/JFMPC

DOI: 10.4103/jfmpc.jfmpc_858_24

How to cite this article: AlMoshary M. Hemorrhagic cholecystitis afflicted with glanzmann thrombasthenia patient. J Family Med Prim Care 2024;13:5395-8.

their potential complications among healthcare providers. This case underscores the critical role of early diagnosis and tailored management strategies in improving patient outcomes by family physicians. By elucidating the complexities of managing such rare conditions, this report advocates for comprehensive, individualized care plans and promotes the integration of specialized knowledge into general practice, ultimately aiming to enhance patient care and prevent adverse health outcomes in similar cases.

Case Presentation

A 27-year-old man with a confirmed diagnosis of Glanzmann thrombasthenia initially visited the emergency department with sudden-onset vertigo and ear pain. The initial evaluation revealed benign paroxysmal positional vertigo (BPPV), which was managed accordingly. However, his next two visits to the emergency department were prompted by episodes of chest pain. Despite electrocardiograms (ECG) showing no signs of ischemic changes, he was given NSAIDs (diclofenac sodium) for presumed musculoskeletal chest pain.

Two days after his initial presentation, the patient returned to the emergency department with intense right upper quadrant abdominal pain radiating to his back, accompanied by nausea and vomiting. A physical examination revealed tenderness, rebound tenderness, and rigidity in the right upper abdominal quadrant. A gastroenterological assessment showed negative Murphy signs but with mild jaundice. Laboratory tests are shown in [Table 1]. An X-ray of the erect abdomen was unremarkable [Figure 1].

Ultrasound imaging revealed a 12 mm diameter echogenic shadowing gallbladder lumen stone associated with extensive sludge formation filling the lumen of the gallbladder, features of acute on top of chronic cholecystitis [Figure 2].

Based on the patient's symptoms, physical examination, and imaging results, we decided to perform an emergency laparoscopic cholecystectomy. During the procedure, we found that the gallbladder was severely inflamed, distended, and had moderate adhesions to the surrounding tissue. Additionally, we observed blood clots in the gallbladder, indicating a hemorrhagic condition. We completed the laparoscopic cholecystectomy. After the procedure, the patient's symptoms significantly improved. We discharged the patient with a prescription for oral medications and scheduled an early follow-up appointment to monitor their progress.

Table 1: Laboratory findings of patients before and after surgery

Laboratory tests	Before surgery	After surgery
Hemoglobin (g/dl)	12.3	11.2
White blood cells (10^3/uL)	12.90	7.42
Neutrophils (%)	20.90	64.4
Platelets (10^3/uL)	274	310
Lymphocytes (%)	14.8	41.90
Total Bilirubin (umol/L)	3.54	1.45
Amylase (ukat/L)	0.6	0.54
Alanine aminotransferase (U/L)	18	19
Creatinine (umol/L)	79.7	75.4
CRP (mg/L)	29.4	34.1
Lipase (U/L)	37.4	37.1
Sodium (mmol/L)	135.4	137.9
Potassium (mmol/L)	3.4	4.1
PT (second)	13.6	13.8
INR	1.01	1.02

Discussion

The emergence of hemorrhagic cholecystitis in a patient diagnosed with Glanzmann thrombasthenia represents a distinctive and scarcely documented pathological association, warranting an in-depth exploration of this rare phenomenon. In the year 1979, Shah and Clegg elucidated the occurrence of hemobilia attributed to cholecystitis, specifically identified as hemorrhagic cholecystitis.[7] Hemorrhagic cholecystitis (HC) is an infrequent occurrence within acute cholecystitis, carrying a significant risk of morbidity and mortality if its management is delayed.[8]

This is the first case of hemorrhagic cholecystitis in a patient with Glanzmann thrombasthenia. The use of NSAIDs for pain management poses a potential contributing factor to the development of hemorrhagic cholecystitis. The pathophysiology involves the antiplatelet effects of NSAIDs, which can exacerbate the inherent bleeding risk in individuals with Glanzmann thrombasthenia.[9,10] These medications inhibit the activity of cyclooxygenase, leading to a decrease in the production of thromboxane A2—a crucial mediator in platelet aggregation.[11,12] Given that Glanzmann thrombasthenia is characterized by defects in platelet integrin αIIbβ3,[1] the additional antiplatelet effects of NSAIDs may further compromise hemostasis and contribute to the occurrence of hemorrhagic cholecystitis.[13] This emphasizes the importance of cautious pain management strategies in individuals with Glanzmann thrombasthenia to

Figure 1: X-ray erect abdomen showing normal gut gases

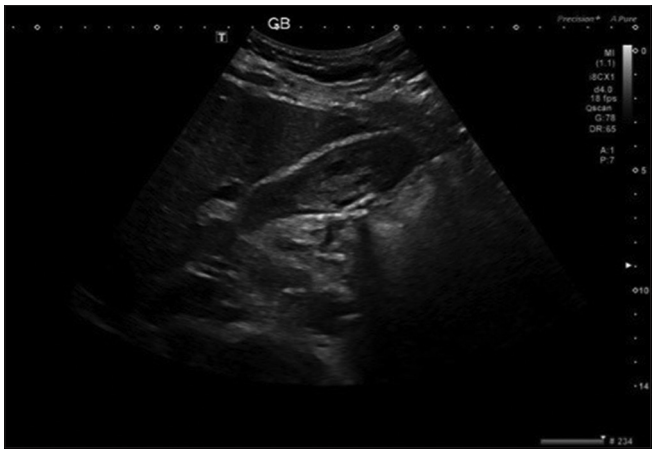

Figure 2: Ultrasound visualization of acute on chronic cholecystitis: echogenic stone, sludge formation, and mural changes in the gallbladder

minimize the risk of complications associated with bleeding, including rare manifestations such as hemorrhagic cholecystitis.

Abdominal ultrasonography (USG) serves as the primary diagnostic tool for patients with acute cholecystitis. However, its effectiveness in diagnosing hemorrhagic cholecystitis remains uncertain. Therefore, the diagnosis is typically established through contrast-enhanced computed tomography (CT). Characteristic CT findings encompass gallbladder wall thickening, gallbladder distension, and the presence of heterogeneous high-attenuation material within the gallbladder lumen.[14,15] In our case CT scan was unremarkable. Additionally, endoscopic retrograde cholangiopancreatography (ERCP) proves beneficial in evaluating hemobilia in select cases.[16,17]

In this case upon surgical exploration, the unexpected revelation emerged that the gallbladder was afflicted with hemorrhagic cholecystitis. This discrepancy underscores the challenge of accurately diagnosing hemorrhagic cholecystitis solely through imaging modalities, emphasizing the imperative role of intraoperative findings to confirm and refine the diagnosis.

Two therapeutic approaches are available for addressing hemorrhagic cholecystitis. The prevailing trend in reported cases involves patients undergoing either open or laparoscopic cholecystectomy, with the latter demonstrating superior outcomes.[5,18] Consistent with this, our case was managed through laparoscopic cholecystectomy, resulting in the patient's successful recovery without encountering any complications.

In individuals with Glanzmann thrombasthenia, it is essential to carefully assess the risks and benefits of various medications. Aspirin and other NSAIDs are contraindicated due to their antiplatelet effects, which can worsen platelet dysfunction. Anticoagulants like Heparin, Warfarin, Ticlopidine, or Clopidogrel require cautious use, as they can affect platelet aggregation. Glycoprotein IIb/IIIa antagonists, such as abciximab, should be used with caution due to the increased risk of bleeding. Thrombolytic agents, including streptokinase, urokinase, or tissue plasminogen activator (tPA), need careful consideration because of their potential to increase bleeding risks. Additionally, volume expanders like dextran or hydroxyethyl starch should be administered with caution to avoid exacerbating bleeding tendencies.[19]

The limitation of this case is the inability to generalize the findings to a broader population due to the singular nature of the case. Moreover, the lack of long-term follow-up data restricts insights into the patient's prognosis and the effectiveness of the treatment.

Conclusion

In conclusion, our case report highlights the rare but critical occurrence of hemorrhagic cholecystitis in a patient with Glanzmann thrombasthenia. This condition necessitates cautious consideration when managing pain with NSAIDs, as the inherent platelet dysfunction poses a significant risk of exacerbating bleeding tendencies. Therefore, healthcare providers should prioritize alternative pain relief modalities and avoid nonsteroidal anti-inflammatory drugs to minimize the likelihood of hemorrhagic complications. A judicious approach to analgesic choices is crucial in Glanzmann thrombasthenia to prevent potentially life-threatening outcomes.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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