Acute hemolysis and methemoglobinemia secondary to fava beans ingestion in a patient with G6PD deficiency
A case report of a rare co-occurrence
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Abstract
Rationale: Favism is a well-known cause of acute hemolytic anemia. Rarely, methemoglobinemia can also happen because of fava bean ingestion in patients with glucose-6-phosphate dehydrogenase (G6PD) deficiency. Few cases with this co-occurrence have been reported in the literature.

Patient concerns: We report a case of a 47-year-old patient who presented with jaundice that started 2 days after eating fava beans.

Diagnoses: Laboratory investigations revealed anemia with evidence of hemolysis (high reticulocytes count, high indirect bilirubin, bite cells in peripheral smear). Blood gases showed high methemoglobin level. Reduced level of G6PD enzyme confirmed the diagnosis of G6PD deficiency.

Intervention: The patient was kept on supplemental oxygen. He was counselled to avoid food and drugs that can cause acute hemolysis.

Outcomes: Oxygen saturation improved gradually. The patient was discharged without any complications after 2 days.

Lessons: Patients with G6PD deficiency can develop both acute hemolytic anemia and methemoglobinemia secondary to fava beans ingestion. These patients should not receive methylene blue to avoid worsening hemolysis.

Abbreviations: G6PD = glucose-6-phosphate dehydrogenase, Hb = hemoglobin, MetHb = methemoglobin, NADPH = nicotinamide adenine dinucleotide phosphate, SatO2 = oxygen saturation.

Keywords: favism, glucose-6-phosphate dehydrogenase deficiency, hemolysis, methemoglobinemia

1. Introduction
Glucose-6-phosphate dehydrogenase (G6PD) deficiency is a common cause of hemolytic anemia with approximately 400 million affected people globally.[1] Patients with G6PD deficiency are typically asymptomatic unless they become exposed to an oxidative stress which induces acute hemolysis. Fava beans ingestion is a well-known factor that can lead to acute hemolysis. Other triggers include infections and certain drugs.[2]

Methemoglobin (MetHb) is an abnormal oxidized form of hemoglobin (Hb) in which the heme iron configuration is changed from ferrous (Fe2+) to ferric (Fe3+) state. Methemoglobinemia is most of the time acquired, resulting from exposure to oxidizing agents such as medications[3] and chemicals. Rarely, methemoglobinemia can be congenital.[4]

In patients with G6PD deficiency, acute hemolysis and methemoglobinemia can happen due to exposure to fava beans. To the best of our knowledge, this co-occurrence is very uncommon and has been described in the literature infrequently. Herein, we report a 47-year-old patient who developed this seldom condition.

2. Case presentation
The patient is a 47-year-old gentleman with a past medical history of hypertension and type-2 diabetes mellitus. He presented to the emergency department with a 3-day history of yellowish discoloration of his eyes and red urine. He denied any history of fever or abdominal pain. He did not complain from dyspnea or dizziness. He is complaint to his home medications and did not start any new medicine recently. He recalled eating a medium-sized plate of fava beans 2 days prior to his symptoms.
In the emergency department, he was febrile 38.5°C with blood pressure of 125/78 mm Hg. His heart rate was 117 beats per minute and respiratory rate was 20 breaths per minute. Oxygen saturation (SatO2) was 88% on room air. Upon examination, he had jaundice. He did not show any sign of distress. The rest of examination was unremarkable. He was put on supplemental oxygen and SatO2 barely increased to 90% to 91%. Blood gases analysis revealed SatO2 of 99% and MetHb level of 3.6% (Table 1).

Complete blood count showed Hb of 12 g/dL (baseline 1 month before was 14.9 g/dL) with high reticulocytes count. Blood chemistry revealed hyperbilirubinemia which was mainly due to high indirect bilirubin (Table 1). We could not get haptoglobin and total bilirubin. Upon examination, he had jaundice. He did not show any sign of distress. The rest of examination was unremarkable. He was put on supplemental oxygen and SatO2 barely increased to 90% to 91%. Blood gases analysis revealed SatO2 of 99% and MetHb level of 3.6% (Table 1).

The patient received paracetamol for the fever, which resolved within 24 hours. He was kept on supplemental oxygen and his SatO2 improved gradually. He did not require blood transfusion and became asymptomatic within 2 days. He was discharged after being counselled about the importance of avoiding food and drugs that may trigger hemolysis. A repeat Hb was done, and the level was low, which confirmed the diagnosis of G6PD deficiency (Table 1).

4. Conclusion

Although it is rare, acute hemolysis and methemoglobinemia can co-occur after fava bean ingestion in patients with G6PD deficiency. Patients with even mildly increased levels of methemoglobinemia may have significant hypoxia and may require hospital admission. Identifying the presence of this co-occurrence is vital as giving methylene blue can worsen the hemolysis.

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Author contributions

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