

CASE REPORT

A Rare Case of Perioperative Methemoglobinemia in a Paediatric Child: A Case Report

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ABSTRACT

Methemoglobinemia is a rare condition that caused impairment of oxygen delivery manifested as spectrum of features ranging from mild desaturations to severe cyanosis and even death. It is rarely seen in the operative theatre and its incidence is scarcely reported. Even low risk patients are susceptible despite having no significant exposure to commonly reported precipitating oxidizing agents. We described a case of perioperative methemoglobinemia in a 10-year-old child, which was planned for foreign body removal in left ear. The operation went successfully; however, the child developed desaturation postoperatively. Arterial blood gas was measured, and methemoglobin levels were noticeably high at 27.1%. The child remained hemodynamically stable, monitored conservatively, and was successfully treated with intravenous methylene blue. We postulated that the cause of methemoglobinemia was intravenous fentanyl usage, as other oxidising agents were excluded. Clinicians must be aware of the possibility of perioperative methemoglobinemia causing unanticipated desaturations and unexplained cyanosis.

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INTRODUCTION

Methemoglobinemia is a rare disorder caused by methemoglobin reductase deficiency at birth or acquired causes including exposure to drugs or substances that cause haemoglobin oxidation. Nitrate derivatives like nitroglycerin and local or topical anaesthetics have been linked to methemoglobinemia and are frequently used in conjunction with anaesthesia. Individuals who are vulnerable to methemoglobinemia, such as those with G-6-PD deficits, renal failure, anaemia, or HIV infection, are more prone to do so when exposed to these oxidising substances. Despite having an oxidising effect, there is no reported incidence of fentanyl linked to methemoglobinemia in the literature (1). Undetected and untreated methemoglobinemia may lead to catastrophic outcomes for patients. Reports on the incidence of methemoglobinemia among paediatric patients detected in the operative theatre are very scarce.

CASE REPORT

This is the case of a 10-year-old Sabahan child who was

admitted to the hospital due to a foreign body, which was a fly insect, in the left ear. The child had no known medical conditions and was planned for an examination of the ear and removal of a foreign body under general anaesthesia. A day before admittance, the child's left ear was purportedly invaded by an insect while she slept. She was otherwise a healthy, active child who weighed 60 kg and had no history of obstructive sleep apnea. There was sufficient time for nil-by-mouth before the general anaesthesia. Prior to the procedure, an airlock assessment revealed that the patient was stable and that all her vital signs were within normal limits. Baseline saturation under room air was recorded at 99%, and a lung exam revealed nothing unusual.

There were no obvious hemodynamic abnormalities while standard monitoring was in place. Both intravenous (IV) propofol (0.5 mg/kg) and IV fentanyl (25 mcg) were given before the supraglottic airway device was inserted. Sevoflurane, a volatile anaesthetic, was used to maintain anaesthesia. Despite having more oxygen at FiO₂ 0.6, a drop in saturation from 99% to 94–96% was noted, and no obvious cyanosis was noticed. The patient was maintained with spontaneous ventilation throughout the procedure. No other drugs or local anaesthetics were used during the procedure other than IV propofol, fentanyl, and volatile anaesthetic agents. Drugs that may be associated with methemoglobinemia

such as eutectic mixture of local anesthetic cream, nitrous oxide, metoclopramide, and paracetamol was also not used in this case. The procedure was successful and took around 20 minutes.

In spite of the decreased saturations, the patient's clinical condition was stable. The patient was extubated in the operating room with 100% oxygen. She was brought to the recovery bay, where a facemask with a 50% oxygen level was used to maintain oxygenation while she was supported in a 30-degree position. During her recovery, despite the supplemental oxygen that was administered, her saturation was noted to be 87-92%. The child appeared cyanotic. There was no chest pain, and a routine lung check revealed an unremarkable breathing rate of about eighteen breaths per minute.

She was subsequently brought into the paediatric surgical critical care unit (PSICU) for close observation and further assessment. Arterial blood gas (ABG) was measured and noted. Acid-base balance and oxygenation was within acceptable limits; however, methemoglobin levels were noticeably high at 27.1%. Therefore, our working diagnosis at that time was hypoxia secondary to methemoglobinemia. Other evaluations, such as a chest x-ray and an echocardiogram, found no obvious pathology. The saturation level was continuously measured, and the paediatric team was consulted for IV methylene blue as treatment for methemoglobinemia. The dose was administered as 1 mg/kg of methylene blue, diluted with glucose 5% as a 1% solution, given intravenously over a 30-minute period as a single dose. The child markedly improved after receiving the methylene blue therapy. Her saturation increased to 98-99% at 40% oxygen. The cyanosis was also resolved. Two hours following the treatment, an ABG was done a second time, and the results showed normal acid-base levels and a significantly decreased methemoglobin level of 2.1%. Vital signs remained stable throughout the stay in the PSICU. The child was transferred to the paediatric ward the next day and discharged two days later.

DISCUSSION

Methemoglobinemia results from the oxidation of the ferrous iron in haemoglobin to the ferric iron state. Methemoglobin cannot carry oxygen; therefore, excessive amounts may interfere with the tissues' ability to receive oxygen. Cyanosis, neurological, and cardiac dysfunction could happen when methemoglobin levels are beyond 30%. Methemoglobinemia, which is linked to a hereditary condition called congenital methemoglobin reductase deficiency, has been reported to cause perioperative cyanosis in some cases (2). In our case, the cyanotic appearance was evident even though the methemoglobin levels were considerably high at 27.1%. Although dietary factors, inherited traits, or even idiopathic causes might contribute to

methemoglobinemia, exposure to oxidising agents is the most common cause of the condition.

According to numerous studies, the most common oxidising agents that put patients at risk for developing methemoglobinemia include local anaesthetics and medications like Dapsone (3). The development of methemoglobinemia is not aided by anaesthetics like propofol and volatile substances like enflurane since they are not oxidants (4). One of the substances that has the potential to render individuals more susceptible to developing methemoglobinemia is fentanyl (1). However, when the literature for this particular topic was sought out, no reports on fentanyl-induced methemoglobinemia were discovered. In our scenario, the patient has no known risk factors for methemoglobinemia. She had never been exposed to or used any of the known oxidising substances over a prolonged period of time. We made an attempt to rule out any potential agents, but the only thing our patient was exposed to during induction was IV fentanyl, which is on the list of drugs that might result in methemoglobinemia. This led us to conclude that the trigger was IV fentanyl.

According to our further assessment, methemoglobinemia is most likely caused by post-exposure to IV fentanyl at induction. The comparatively low but sufficient IV fentanyl dose administered may be responsible for the initial desaturation of 94-96%, which was not as severe intraoperatively. However, because the patient might have had unidentified underlying genetic disorders such as methemoglobin reductase deficiency, we were unable to definitively prove that IV fentanyl was the primary cause. Evaluation of MetHb, CYB5R activity assessment, and genotyping are the main diagnostic assays in the differential diagnosis. The most accurate method for determining MetHb concentrations is by applying the change in MetHb absorbance at 630 nm that occurs when cyanide is introduced, turning MetHb into cyan-MetHb (5).

Despite the fact that there aren't any certain risk factors for methemoglobinemia, especially in this young child, this case highlights the significance of comprehending the potential interactions between fentanyl and methemoglobinemia. Methemoglobinemia may develop among individuals with unidentified genetic diseases, especially in those who may be having their first anaesthetic experience. The patient's parents declined to consent to further genetic testing or medical investigation, despite our eagerness to do so for this child. Therefore, that limitation will provide a starting point for the additional research required to further clarify the prevalence of genetically related methemoglobinemia in distinct Sabah State regions.

CONCLUSION

Unaccounted-for hypoxia during the recovery period

from surgery should alert medical professionals to the possibility of methemoglobinemia, and prompt examination and treatment should follow. Clinicians should be mindful that medicines used in the operating room, which may not have been reported previously, can also cause methemoglobinemia, as we hypothesized in this case with the use of fentanyl. Although propofol and volatile anesthetic drugs do not have oxidizing properties, their prospective interactions with vulnerable individuals in potentially causing methemoglobinemia warrant more investigation. This case also raised the question of genetic susceptibility amongst Sabahan ethnicity to this condition, although its exact incidence has not been reported previously. This could be a potential genetic research area in the future.

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