CASE REPORT

The painted shoes

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SUMMARY

A previously well 4-year-old boy presented to the emergency room with progressive cyanosis, pallor and vomiting over the last 5 h. Oxygen saturation on pulse oximetry was 87–89% despite 9 L/min of supplemental oxygen. He was tachypnoeic and had a systolic heart murmur, with no other findings on clinical examination. In his medical history, there was record of a restrictive atrial septal defect, with a normal echocardiogram from 3 years before. He had no relevant family history. His shoes appeared to have been recently painted, which raised the suspicion of methaemoglobinaemia, presumptively caused by aniline-containing shoe dye. The shoes were removed promptly and his feet washed profusely. After confirming the diagnosis, methylene blue was started. The level of methaemoglobin decreased rapidly and the boy made a full recovery.

BACKGROUND

Cyanosis is a condition that may result from a variety of situations, some of which are life-threatening. After excluding respiratory and circulatory causes, it is important to consider other less frequent diagnoses and begin treatment promptly. We would like to draw the attention of the reader to the importance of a thorough clinical history and physical examination, which, in this case, were the key to the diagnosis.

CASE PRESENTATION

A previously healthy 4-year-old boy was admitted to the emergency room (ER) with cyanosis, pallor and vomiting. The cyanosis and pallor were progressive and noticed during a religious procession (figure 1) 5 h before admission, with no other accompanying manifestations. About 30 min before being admitted to the ER, the boy had started vomiting.

The child had a history of restrictive atrial septal defect, with a normal echocardiogram from 3 years before. He had no relevant family history.

On clinical examination, he was sleepy but conscious and reactive. He had central and peripheral cyanosis (figure 2) as well as pallor of the skin. He was tachypnoeic, with normal breath sounds, and his oxygen saturation on pulse oximetry was 87–89% despite 9 L/min of supplemental oxygen. He was non-feverish, and his blood pressure was 118/83 mm Hg and pulse rate 124 bpm. On cardiac auscultation, the patient had a systolic murmur grade I-II/VI, more audible on the left sternal border. The rest of the examination was normal.

On further observation, we noticed that the boy's black shoes appeared to have been recently

Figure 1 Lip cyanosis during the religious procession.

painted (figure 3). After asking the mother, she confirmed the shoes were previously white and had been painted on that day. The child was wearing thin lacy socks and had put on the shoes 1 h before

Figure 2 Skin pallor and central cyanosis in the emergency room.

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Figure 3 Recently painted shoes.

the symptoms had started. There were dye stains inside the shoes, on the thin lacy socks and also on the child's feet.

INVESTIGATIONS

An arterial blood sample was taken and showed a chocolate-brown colour (figure 4). Owing to a technical problem, the blood gas analysis of this sample could not be performed. Venous blood gas analysis showed pH 7.22, pCO₂ 60 mm Hg, pO₂ 18 mm Hg and O₂ saturation of 18% with methaemoglobin (MetHb) concentration of 24.1% (reference range 1–1.5%).

DIFFERENTIAL DIAGNOSIS

The patient had a normal pulmonary auscultation and no signs of respiratory distress other than tachypnoea and cyanosis. He had a previously known functional heart murmur and no signs of haemodynamic compromise besides pallor. After excluding the most common causes of cyanosis—respiratory and cardiac—other less frequent diagnoses were considered. The shiny black shoes caught our attention and we equated the diagnosis of methaemoglobinaemia, presumptively due to aniline-containing shoe dye.

TREATMENT

When methaemoglobinaemia was suspected, the shoes and socks were immediately removed and the boy's feet were

Figure 4 Chocolate-brown discolouration of the arterial blood.

washed. After laboratory confirmation, he was started on intravenous methylene blue, 1–2 mg/kg of 1% solution, administered over 5 min.

Shortly after the administration of methylene blue, the peripheral saturation of O₂ started to improve and the cyanosis disappeared. The concentration of MetHb dropped to 1.8% 2 h later.

OUTCOME AND FOLLOW-UP

The patient was kept under observation until his MetHb level was 0%. He was discharged 18 h after admission, with a peripheral oxygen saturation of >97%.

Two weeks after being discharged, he was reassessed in the haematology outpatient clinic to exclude the existence of a genetic susceptibility to the disease. Glucose-6-phosphate dehydrogenase deficiency, haemoglobinopathies (by high-performance liquid chromatography) and changes in cytochrome B5 reductase, were ruled out.

The shoemaker was contacted. He admitted having used an alcohol-based shoe dye containing aniline (confirmed in the label), instead of the regular water-based dye (without aniline), as the former dries faster and the shoes had to be ready on that same day for the procession.

DISCUSSION

Cyanosis is a bluish discolouration of the tissues near the skin surface due to an increased concentration of deoxygenated haemoglobin (Hb) in capillary blood. There are two main types of cyanosis—central and peripheral. Central cyanosis is usually due to systemic arterial oxygen desaturation, while peripheral cyanosis generally results from increased oxygen extraction by the tissues.¹

After excluding the most common causes of cyanosis, the major being respiratory and circulatory, other less frequent but still life-threatening conditions must be considered.

Methaemoglobinaemia should be suspected based on the clinical history, physical examination and on discrepancy between the oxygen saturation obtained by routine pulse oximetry and the oxygen saturation calculated from arterial blood gas analysis—'saturation gap'.^{2–3} In this case, a thorough physical examination raised the suspicion of methaemoglobinaemia and the patient was treated accordingly in a short time frame despite the fact that the partial pressure of arterial oxygen (PaO₂) could not be determined, as previously mentioned.

Aniline and its derivatives are highly toxic oxidant compounds used in the manufacture of precursors to polyurethane, dyes, rubber, herbicides and other industrial chemicals. The exposure to these compounds may occur via oral ingestion and absorption through the lungs or skin, and may result in clinically significant methaemoglobinaemia, depending on the concentration of MetHb in the blood.^{4–5} In our case, there were dye stains inside the patient's shoes, and on his socks and feet, suggesting cutaneous absorption.

MetHb is a modified form of Hb, in which the ferrous ion (Fe²⁺) is oxidised to the ferric state (Fe³⁺). Ferric ions are not able to bind oxygen. In the presence of methaemoglobinaemia, the ferrous haemes of the remaining Hb molecules have increased affinity for oxygen. This results in a 'left shift' of the oxygen dissociation curve, with impaired delivery of oxygen to the tissues.

In normal circumstances, MetHb levels are less than 1%,⁵ due to endogenous mechanisms that protect against oxidative stress. Nevertheless, the exposure to an oxidising agent, such as aniline, or to its metabolites, increases the conversion of Hb to MetHb, which may overwhelm these protective mechanisms.^{2–6}

Treatment of patients with methaemoglobinaemia depends on the severity of the disorder.^{2 4–6} For lesser degrees of MetHb (<20%), there is generally no therapy required other than discontinuation of the offending agent(s). Specific therapy with methylene blue is indicated for patients whose level of MetHb is higher than 20% or for those who are symptomatic.^{4 7} Methylene blue provides an artificial electron transporter for the ultimate reduction of MetHb to Hb.^{6 7}

As a potentially fatal condition, it is imperative to diagnose it promptly and to initiate treatment as required.

Learning points

- ▶ Methaemoglobinaemia should be considered in the presence of cyanosis with no apparent respiratory or cardiac cause.
- ▶ Methaemoglobinaemia should be suspected when there is a 'saturation gap'.
- ▶ A detailed clinical history and thorough physical examination may be the key to the diagnosis.
- ▶ Methylene blue is indicated in the treatment of patients with methaemoglobinaemia whose level of methaemoglobin is higher than 20% or in those who are symptomatic.

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