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Clinical Picture

Rachel Bone

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Black oesophagus in an adolescent with type 2 diabetes

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A 13-year-old boy with a past medical history of poorly controlled type 2 diabetes and obesity was admitted with massive haematemesis, melaena, and severe epigastric pain. He had developed non-autoimmune diabetes (glutamate decarboxylase antibodies, islet tyrosine phosphatase 2 antibodies , insulin autoantibodies, and Zinc Transporter 8 antibodies were absent); HbA_{1c} 14·5% at the age of 10 years, having multiple paternal inheritance. Although known to have a bulimic behaviour, his medical history was negative for gastrointestinal disorders and for gastrointestinal toxic drugs or caustic consumption.

At admission, he had very poor metabolic control (glycaemia 840 mg/dL, HbA_{1c} higher than 13%) because he had voluntarily withdrawn insulin and refused metformin. He complained of polyuria and polydipsia for several days before admission, had an altered mental status, and extreme dehydration of both skin and mucous membranes with a prolonged capillary refill (>2 sec). The initial blood count was 6 130 000 red cells per mL, haemoglobin 18·8 g/dL, haematocrit 54·5%, and 555 000 platelets per mL. Blood biochemistry analyses showed ketoacidosis (pH 7·07, HCO3 9·7 mmol/L, and blood ketones >7 mg/dL) and stage 3 prerenal acute kidney injury. Inflammatory indexes and blood toxicology screening were negative. His clinical condition was rapidly deteriorating and he was extremely dehydrated, in shock, tachycardic, unresponsive to verbal stimuli, and developed Kussmaul breathing. Soon after massive haematemesis episodes, blood tests showed severe normochromic anaemia (haemoglobin 7·4 g/dL [reference 13–16 g/dL])

The endoscopic examination after removal of abundant blood residues showed circumferential black oesophagus appearance, sparing only the first proximal 3–4 cm (figure 1A). The stomach was filled with blood material and several mobile clots. After thorough cleansing, no sources of active or recent bleeding were found in the stomach or the duodenum. No biopsy samples were collected to avoid the risk of perforation. A CT angiogram showed oesophageal wall thickening and millimetric gaseous microareolas in pneumomediastinum (gas bubbles smaller than 1 cm, indicating esophageal microperforation).

The patient was treated with exclusive parenteral nutrition with continuous insulin infusion and monitoring of glycaemia, proton-pump inhibitors infusion, oral sucralfate, intravenous fluconazole, and broad antibiotic intravenous coverage. Within 4 weeks, the epigastric pain had resolved and the patient was able to feed orally safely. A repeated oesophagogastroduodenoscopy 1 month after admission showed complete recovery of the oesophageal mucosa, except for the presence of mild circumferential scar striae (figure 1B; green arrow). Findings on CT angiogram of the chest and abdomen were normal.

Acute oesophageal necrosis is a rare clinical entity of unknown cause that has been reported in the setting of multiorgan dysfunction, sepsis, diabetic ketoacidosis, thromboembolic disorders, alcohol intoxication, gastric volvulus, and cancer. Men are four times more affected than women and the peak incidence occurs at an average age of 67 years. A mortality rate of 7% has been reported, mainly linked to underlying diseases. To the best of our knowledge, this case is the first instance of so-called black oesophagus in paediatric age, perhaps an intriguing complication of childhood-onset diabetes. From the possible comorbidities, our patient had only diabetic ketoacidosis. Therefore, we hypothesise that blood hypo—perfusion due to the extreme dehydration linked with ketoacidosis was the main factor leading to oesophageal ischaemia. Paediatricians addressing haematemesis in children with the aforementioned underlying conditions should be aware of oesophageal ischaemia as a possible differential diagnosis.

Contributors

All authors were involved in the care of the patient, in the writing of the manuscript, and in the decision to submit for publication. Written informed consent was obtained from the patient's parents for the publication of both clinical information and imaging.

Declaration of interests

We declare no competing interests.

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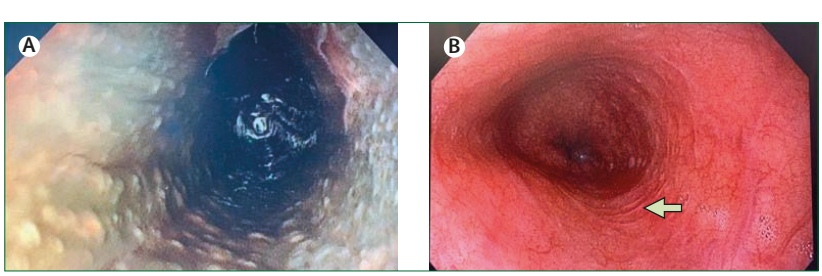

Figure: **Esophageal endoscopic images**
(A) Black oesophagus in a 13-year-old boy with type 2 diabetes. (B) Complete recovery of the oesophageal mucosa, except for mild circumferential scar striae (green arrow), after 1 month of treatment.