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CASE REPORT

Boerhaave Syndrome in a Paediatric Patient With Diabetic Ketoacidosis: A Case Report

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Abstract

Diabetic ketoacidosis is a serious and potentially life-threatening complication of type 1 diabetes mellitus (T1DM), which often results from poor glycaemic control. While diabetic ketoacidosis is well documented, rare complications such as Boerhaave syndrome can considerably increase morbidity. This case report presents a 14-year-old girl with poorly controlled T1DM and celiac disease who was admitted with diabetic ketoacidosis and developed severe vomiting and abdominal pain. Imaging studies revealed the presence of pneumomediastinum, which raised concerns about oesophageal injury and a diagnosis of Boerhaave syndrome was made. The patient was managed conservatively with correction of metabolic acidosis, intravenous antibiotics, supportive care and close monitoring. Her condition improved gradually, with resolution of ketoacidosis and stabilisation of the pneumomediastinum without invasive intervention. This case highlights the importance of early identification and the adoption of a multidisciplinary approach in managing atypical complications of diabetic ketoacidosis. Timely intervention can ensure favourable outcomes even in the presence of severe complications.

Key words: Diabetic ketoacidosis; Boerhaave syndrome; Diabetes mellitus, type 1; Paediatrics; Emergencies; Vomiting.

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Introduction

Diabetic ketoacidosis (DKA) is a severe and potentially life-threatening complication of type 1 diabetes mellitus (T1DM) and is characterised by hyperglycaemia, ketosis and metabolic acidosis. The management of DKA in paediatric patients requires careful monitoring and prompt intervention to prevent morbidity and mortality. Common complications include cerebral oedema, hypokalaemia, hypophosphatemia and acute kidney injury.[1] In addition to these common complications, DKA can occasionally result in other rare but serious conditions such as rhabdomyolysis, thrombosis, pulmonary oedema and acute respiratory distress syndrome.[1, 2] Boerhaave syndrome is a rare but serious condition characterised by a spontaneous oesophageal rupture, often precipitated by severe vomiting. A few cases of Boerhaave syndrome occurring in association with DKA have also been reported in the literature.[3-5]

This report presents the first documented case from Saudi Arabia describing DKA complicated by Boerhaave syndrome in a child.

Case history

A 14-year-old girl presented to the emergency department (ED) with vomiting, poor oral intake and abdominal pain. The onset of vomiting was sudden and occurred 5–6 times per day. The volume was high and it was non-bilious and non-projectile. The abdominal pain was mild, localised to the epigastric region, non-radiating and lacked specific aggravating or relieving factors. The patient had a history of T1DM and was diagnosed at 9 years of age. She was on a basal-bolus insulin regimen with insulin degludec (44 units) and insulin aspart (16–20 units before meals). Her diabetes management was suboptimal, as indicated by elevated haemoglobin A_1c levels of 10.4 mmol/L. Additionally, she had celiac disease and had not adhered to a strict gluten-free diet. Her history also included vitamin D deficiency for which she was receiving treatment and a thyroid nodule that was under regular follow-up with a normal recent thyroid stimulating hormone (TSH) level.

On examination, the patient appeared acutely ill and exhibited signs of severe dehydration, such as dry mucous membranes, decreased skin turgor and moderate epigastric tenderness. Vital signs revealed a tachycardia (heart rate 106/min), blood pressure of 112/64 mm Hg and SpO_2 of 98 % on room air. Head and neck examination revealed a congested throat. Furthermore, abdominal examination indicated epigastric tenderness with a soft, non-distended abdomen and normal bowel sounds were present. Respiratory assessment showed good bilateral air entry, with scattered wheezes and rhonchi. The rest of the examination was normal.

When the patient arrived at the ED, her blood glucose level was considerably elevated at 16.6 mmol/L (300 mg/dL) and initial blood gas analysis showed a pH of 7.142, bicarbonate (HCO_3-) of 9.6 mEq/L and a base excess of –24 with positive serum ketones of 2.9 mmol/L. Her white blood cell count was 14.5 x 10^9/L with 70 % neutrophils and 22 % lymphocytes and her C-reactive protein was 22.5 mg/L.

She was admitted to the paediatric intensive care unit (PICU) for intravenous rehydration, insulin infusion, further investigations and close monitoring. Vital signs, hydration status, glucose levels, blood gases, electrolyte imbalances and ketones were meticulously monitored.

Her clinical status improved gradually with insulin therapy and correction of dehydration. However, severe vomiting and abdominal pain persisted, which raised concerns about possible complications. X-rays of the abdomen revealed diminished bowel gases and the colon was loaded with faecal material. A computed tomography

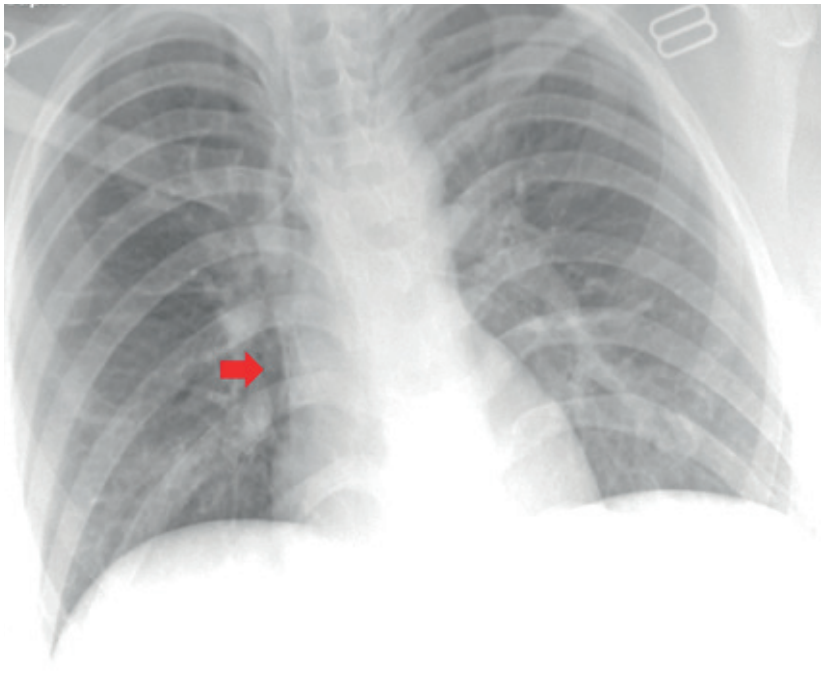

Figure 1: Chest posterior-anterior (PA) X-ray showing linear lucency (arrow) involving the mediastinum and right heart border concerning pneumomediastinum

(CT) scan of the abdomen and pelvis with contrast enhancement did not show any signs of acute abdominal issues. However, the visualised parts of the lower chest demonstrated the presence of pneumopericardium and a chest X-ray confirmed spontaneous pneumomediastinum and pneumopericardium (Figure 1).

A CT chest scan was then done that showed the pneumomediastinum extended to the neck, with air displacing the left posteromedial lung pleura and surrounding the oesophagus, which raised concerns about possible oesophageal injury (Figure 2).

Upon reassessment, the patient was observed to have mild neck swelling with crepitations. A paediatric gastroenterologist and a paediatric surgeon were consulted and a diagnosis of Boerhaave syndrome was made. A plan was devised to manage the patient with close observation and supportive care. A multidisciplinary approach that included input from endocrinology, gastroenterology and paediatric surgery was essential for managing potential complications. The treatment was focused on insulin therapy and correction of metabolic acidosis and electrolyte imbalance. The pneumomediastinum was managed conservatively, the patient was kept *nil* by mouth, piperacillin–tazobactam and antifungal therapy were started and she was watched closely for respiratory distress or signs of mediastinitis.

During her hospital stay, her blood glucose levels normalised gradually with insulin therapy and metabolic acidosis resolved. The anion gap decreased, which indicated the resolution of ketoacidosis. Serum lactate levels normalised and electrolyte imbalances were corrected. A follow-up chest CT scan with oral contrast demonstrated interval improvement in the pneumomediastinum, with no evidence of oesophageal contrast leakage and only minimal pericardial effusion. (Figure 3). A barium swallow test revealed the absence of definite contrast extravasation (Figure 4).

Clinically, her vomiting subsided and she was able to resume oral intake. Abdominal pain lessened and respiratory symptoms resolved without invasive intervention.

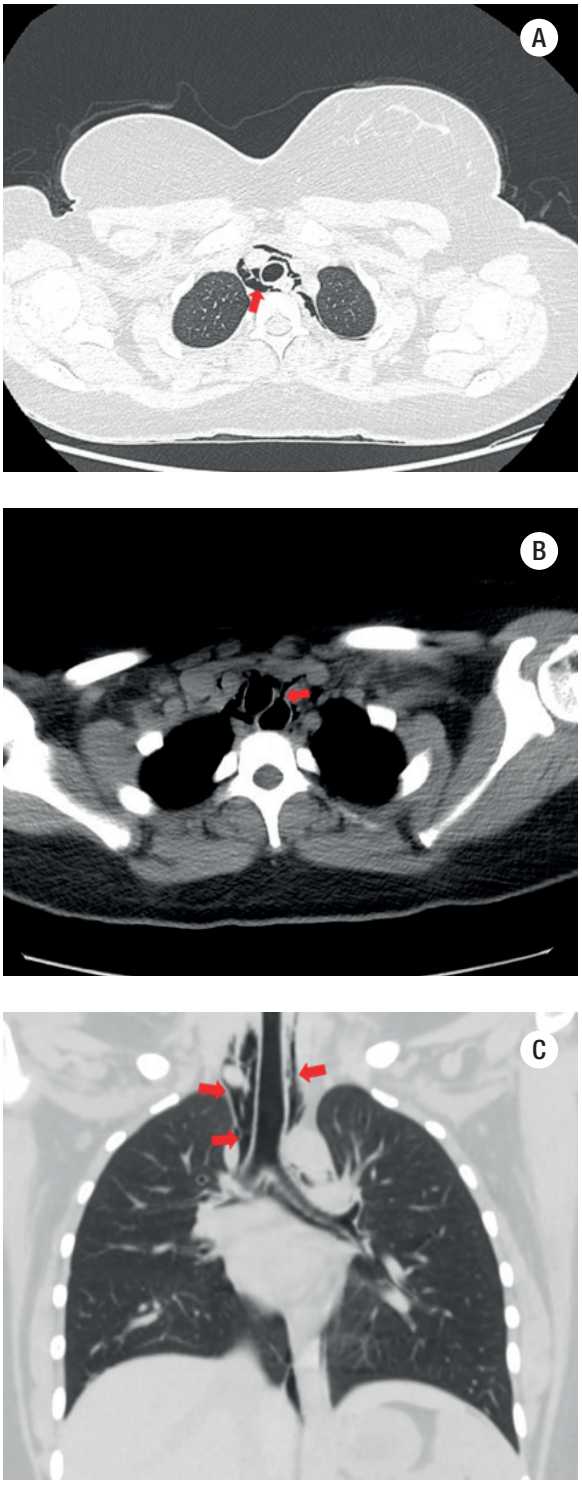

Figure 2: A: Chest computed tomography (CT) scan axial cut showing pneumomediastinum surrounding the oesophagus; B: Chest CT scan axial cut showing a defect in the anterior oesophagus concerning perforation; C: Chest CT scan coronal cut showing pneumomediastinum extending to the neck;

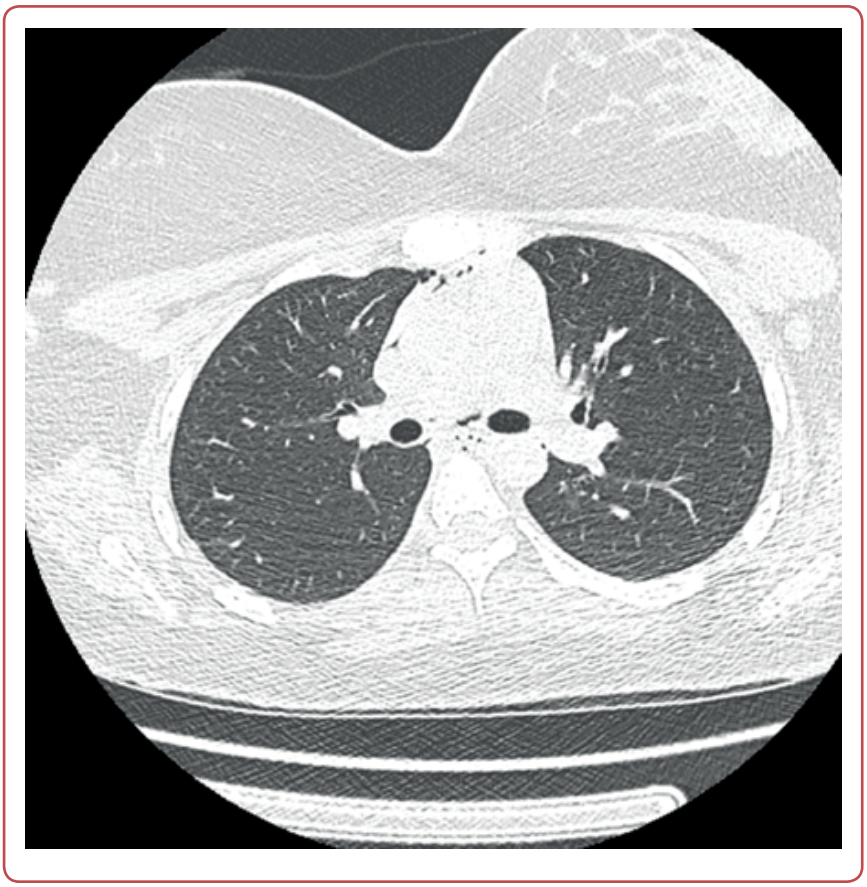

Figure 3: Chest computed tomography (CT) scan axial cut showing regression of pneumomediastinum

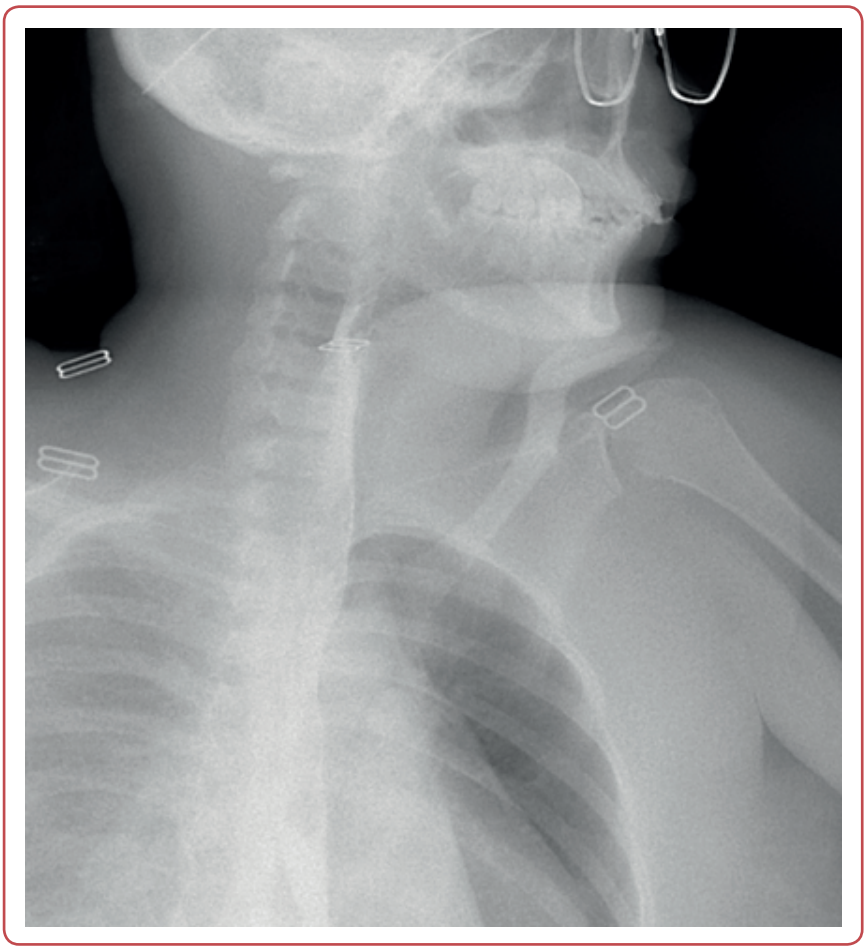

Figure 4: Barium swallow showing no contrast leakage

By discharge, her laboratory parameters were normal and she was stable. Outpatient follow-up with endocrinology, gastroenterology and paediatric surgery was recommended to support ongoing management and prevent complications.

Discussion

Boerhaave syndrome is a rare but serious condition characterised by a full-thickness rupture of the oesophagus and is caused by a sudden increase in intraoesophageal pressure, often due to forceful vomiting.[4] The condition most commonly affects the distal oesophagus, particularly the posterolateral aspect, where the oesophageal wall is more vulnerable owing to the relative lack of longitudinal muscle fibres and an increased concentration of neurovascular structures.[6] The rupture usually occurs in the left pleural cavity, leading to potentially life-threatening complications such as mediastinitis, empyema, or necrosis because of the entry of gastric contents into the mediastinal space.[4, 6] The incidence of Boerhaave syndrome is approximately 3.1 per 1,000,000 people annually, with most cases occurring in middle-aged men. Therefore, it is exceedingly rare in paediatric patients, particularly in those with coexisting conditions such as DKA.[6]

Boerhaave syndrome routinely presents with the Mackler triad of vomiting, lower thoracic pain and subcutaneous emphysema, although this classic presentation is rare.[6] Symptoms are more often nonspecific and may include chest, neck and abdominal pain, odynophagia, dysphagia, vomiting, haematemesis and respiratory distress, with physical signs such as subcutaneous crepitation, the Hamman sign and tachycardia.[4, 6, 7] In the context of DKA, these symptoms can become more complex. For example, Wiggins et al reported persistent vomiting and diffuse abdominal pain in a 22-year-old man,[4] whereas Mitchell and Jones observed severe neck pain and surgical emphysema in a 30-year-old man.[3] Furthermore, Khan et al described a 16-year-old girl with vomiting and subcutaneous emphysema.[8] In contrast, our 14-year-old patient presented with severe abdominal pain and vomiting.

Presented case is noteworthy because of the patient's young age and the simultaneous occurrence of DKA and Boerhaave syndrome, an extremely rare combination. Pneumomediastinum, a complication associated with DKA, occurs when air accumulates in the mediastinal space owing to the rupture of alveoli or the tracheobronchial tree.[9] In DKA, this accumulation can result from elevated intrathoracic pressure, which is often induced by severe and sustained vomiting or coughing.[6, 9] However, in presented patient, pneumomediastinum was likely secondary to Boerhaave syndrome rather than a distinct complication of DKA as she presented with severe gastrointestinal symptoms and vomiting but lacked substantial respiratory symptoms such as cough or dyspnoea.

The diagnosis of Boerhaave syndrome is often challenging owing to its nonspecific presentation. Imaging studies, including chest X-rays and CT scans are critical in detecting complications such as pneumomediastinum, pneumopericardium and subcutaneous emphysema. Although endoscopy is beneficial in visualising the oesophageal tear, it should be used with caution because of the risk of exacerbating the injury.[3, 4, 6, 9] In presented case, a chest CT scan confirmed the presence of pneumomediastinum, which, along with the patient's clinical presentation, raised the suspicion of oesophageal rupture.

Management of Boerhaave syndrome depends on the extent of the rupture, the patient's condition, and the presence of mediastinitis. Early diagnosis is critical as delayed treatment is associated with increased mortality, particularly in those with comorbid conditions.[8] In stable patients without mediastinitis, conservative management, as adopted in this case, may be appropriate. This approach includes fasting, broad-spectrum antibiotics, proton pump inhibitors, fluid resuscitation and parenteral impression as well as close monitoring for signs of deterioration or the need for surgical intervention.[3, 4]

The prognosis of Boerhaave syndrome in the context of DKA largely depends on timely diagnosis and management, as delayed treatment significantly increases morbidity and mortality. Favourable outcomes with conservative management have been documented; for instance, Mitchell and Jones reported a 30-year-old man who recovered without major complications and Khan et al described a 16-year-old girl who achieved full recovery within eight days of conservative care.[3, 8] In contrast, Wiggins et al reported a 22-year-old man who was transferred to a ter-

tiary facility but was lost to follow-up after initial conservative treatment.[4] These cases, along with presented, suggest that with prompt and appropriate conservative management, prognosis can be excellent even in the presence of DKA. However, careful monitoring and a multidisciplinary approach are critical, as complications such as mediastinitis or sepsis can severely impact outcomes if not promptly addressed.[6]

Conclusion

Boerhaave syndrome is a rare but serious complication of DKA that requires prompt recognition and intervention to prevent life-threatening outcomes. This case underscores the importance of considering atypical complications in paediatric DKA and highlights the value of a multidisciplinary approach in managing complex cases. Ongoing documentation of similar cases will help further refine management strategies and improve understanding of this rare association.

Ethics

Our institution does not require ethics approval for reporting individual cases or case series. A written informed consent for anonymised patient information to be published in this article was obtained from the patient's legal guardian.

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None.

Conflicts of interest

The author declares that there is no conflict of interest.

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Data access

The data that support the findings of this study are available from the corresponding author upon reasonable individual request.

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Conceptualisation: YA
Formal analysis: YA
Investigation: YA
Data curation: YA
Writing - original draft: YA
Writing - review and editing: YA
Visualisation: YA

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