CASE REPORT

Cardiopulmonary complications in a patient with bezoar

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SUMMARY

We report a case of a 17-year-old institutionalised male with a medical history of fragile X syndrome, bilateral congenital glaucoma, cataracts and pica disorder. He was transferred to our paediatric intensive care unit owing to respiratory failure and hypotension. On transoesophageal echocardiogram, he presented left atrium compression. A CT of the thorax and mediastinum revealed an unknown heterogeneous material in the lumen of the stomach and oesophagus, with a lung parenchyma suggestive of alveolar foreign material. Endoscopic evaluation showed diaper fragments inside the oesophagus and stomach. Fragmentation and suction of diaper material was made. Medical treatment was performed with inotropic support, conventional mechanical ventilation and antibiotics.

BACKGROUND

The ingestion of foreign bodies can cause severe complications.[1] This is particularly true in children with pica who are institutionalised. Pica is the most dangerous form of self-injurious behaviour exhibited by people with developmental disabilities, and can result in death and other life-threatening events.[2]

Bezoars are classified according to their composition. The most common types are trichobezoars (ingested hair), phytobezoars (vegetable matter) and pharmacobezoars.[3] Less commonly, lythobezoars (stones in the digestive tract) and a variety of other substances like paper or tissue can be found.[4–8] Although the prevalence of bezoar formation is low, the mortality can reach 30% if no treatment is administered, primarily because of gastrointestinal bleeding or perforation.[9]

CASE PRESENTATION

This patient was a 17-year-old boy institutionalised adolescent, with a medical history of fragile X syndrome, mental retardation, bilateral congenital glaucoma, cataracts and pica disorder with reference of eating diapers.

Suddenly he started with respiratory distress and stridor. At arrival of the emergency team, he was unconscious, pale, with hypotension and respiratory distress. Oxygen saturation was of 85% on room air. He was intubated and transferred to our paediatric intensive care unit (PICU).

Fluid resuscitation and inotropic support with dopamine were initiated. Owing to persistent hypotension, a transthoracic echocardiogram was performed and revealed a left atrium compression.

INVESTIGATIONS

Initial capillary blood gases analyses showed a metabolic acidosis with pH 7.21, pCO_2 50 mm Hg, pO_2 50 mm Hg, bicarbonate 20 mEq/L and base excess −8 mEq/L, with a lactate of 5 mmol/L. His haematological blood tests showed a haemoglobin level of 13.6 g/dL, leukocytosis 22 000 cells/μL with 92% neutrophils, platelets counts of 302 000 cells/μL. The biochemical tests were normal. The C reactive protein was negative (0.89 mg/dL). On entrance at the PICU, arterial blood gases showed a pH 7.35, $paCO_2$ 40 mm Hg, paO_2 109 mm Hg, bicarbonate of 20 mEq/L, base excess of −3.8 mEq/L and lactate of 3 mmol/L.

Chest radiography showed bilateral interstitial infiltrate, an enlarged mediastinum and a gastric chamber with normal air content.

Transthoracic and a transoesophageal echocardiography showed a normal diastolic and systolic function with a small left atrium compressed by a mass highly suspicious of being extrinsic to the heart (figure 1).

A thoracic and mediastinal CT showed severe gastric and oesophageal distension with unknown heterogeneous material (figures 2 and 3), diffuse lung parenchyma infiltrates (especially in the left inferior lobe) suggestive of alveolar filling by foreign material, and normal cardiac cavities.

An upper endoscopy detected oesophageal filling by synthetic material (diapers) together with silica crystals (figure 4). In the stomach, there was a tightly packed collection of partially digested food and undigested synthetic material that was stuck in the stomach (gastric bezoar).

A rigid bronchoscopy showed fragments of food in the airways, but no rests of diaper-related material was found.

DIFFERENTIAL DIAGNOSIS

In a previous asymptomatic adolescent, with a history of pica and a sudden onset stridor with respiratory distress, foreign body aspiration was the first diagnostic hypothesis. This situation is more common in children than adults, especially in those younger than 3 years.[10 11] In adolescents and adults, neurological impairment, alcohol or sedative abuse and psychiatric disorders are risk factors for foreign body aspiration.

Although extrinsic left atrium compression is rare, it can be caused by mediastinic (bronchogenic cysts, mediastinal lymphomas and carcinomas), gastrointestinal, (gastric or oesophageal distension, achalasia, diaphragmatic hernia, oesophageal leiomyosarcoma), pulmonary (lung tumour, bronchogenic cysts), aorta

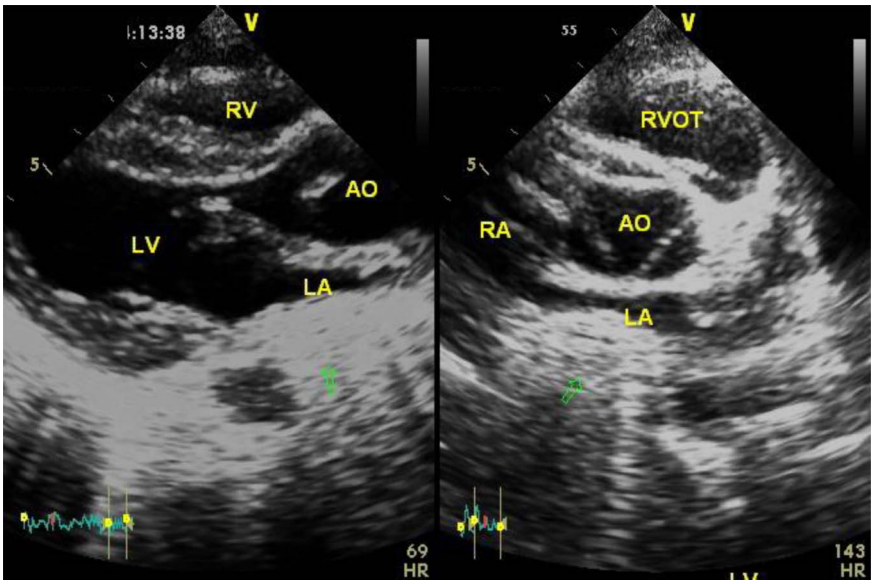

Figure 1 Transoesophageal echocardiogram showing left atrium compression (green arrows). LV, left ventricle; LA, left atrium; RV, right ventricle; Ao, aorta and RVOT, right ventricular outflow tract.

and intrapericardial structures (thoracic aorta aneurysms, aortic root dilatation).[12] Symptoms of left atrium compression include dyspnoea, exercise intolerance. In extreme cases, haemodynamic instability and pulmonary oedema can be present. Owing to poor definition of the mass, intracardiac tumours were also considered in this case. Although rare, most intracardiac tumours are benign. In children, the rabdomyosarcoma is the most frequently found followed by fibroma and teratoma. In adults, atrial myxomas are most frequently found especially in left atrium where they can cause symptoms of heart failure, mitral valve regurgitation or pulmonary hypertension. Primary malignant cardiac tumours (lymphoma; sarcoma) are present in 10% of the cases. A metastatic lesion is even rarer and generally involves the myocardium and pericardium.

An infectious process like bacterial tracheitis, epiglottitis or viral croup, should also be considered in a case of stridor. However, in this particular example, a previous history of fever, ill appearance, hoarseness and upper respiratory symptoms were not reported. Severe allergic reactions (anaphylaxis and laryngeal oedema) can also be considered with abrupt and severe onset stridor.

Figure 2 A thoracic CT showing a heterogeneous material in the stomach.

Chronic or recurrent episodes of stridor should suggest an exogenous compression of the airway caused by a foreign body, subglottic stenosis, vascular rings or a tumour.

TREATMENT

Partial removal of the foreign material present in the oesophageal lumen was performed. It was not possible to remove the bezoar from the stomach. After discussion with paediatric surgeons and paediatric gastroenterologists, it was decided not to perform a surgical intervention of the gastric bezoar and to follow-up on a 2-weekly basis. Bronchial toilet was performed during bronchoscopy.

Inotropic support was reduced and he was extubated on day six of admission. He was treated for 10 days with amoxicillin/clavulanate and gentamicin. An echocardiogram was performed before hospital discharge which showed normal left atrium.

OUTCOME AND FOLLOW-UP

An abdominal ultrasound on day 7 did not show gastric distension. He was eating normally and never presented signs of intestinal obstruction.

DISCUSSION

Pica is a disorder characterised by perverted appetite for substances not fit as food or with no nutritional value. The word 'pica' is derived from Latin word for Magpie, a species of bird that feeds on whatever it encounters. The diagnosis requires that the patient is persistently eating non-food substances for at least 1 month and such behaviour is inappropriate for the child's stage of development.[13] There is a 25% incidence of pica among the developmental delayed population.[14] The aetiology of pica is not known; it has been suggested that may be a consequence of parental neglect and deprivation early in life.[4]

Bezoars develop after the ingestion of foreign bodies that accumulates in the gastrointestinal tract because of large particle size, indigestibility, gastric obstruction or intestinal stasis.[9] Trichobezoars are more frequently seen in young people, and usually result from an underlying behavioural disorder.[9] Clinically, they remain asymptomatic for many years, and have insidious complaints, generally related with their location, including abdominal pain, nausea, vomiting, early satiety, anorexia and weight loss.[15]

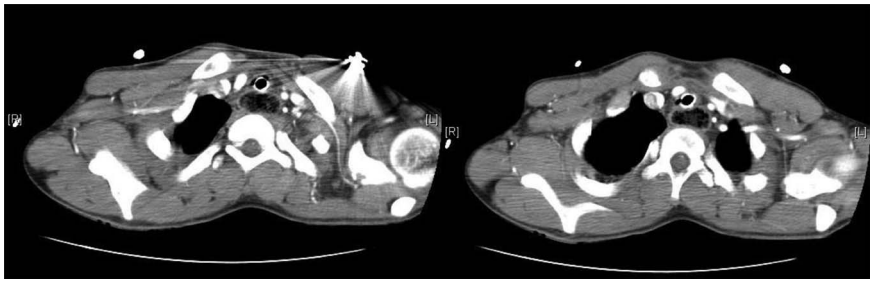

Figure 3 A thoracic CT showing oesophageal distension by a heterogeneous material.

Complications have been described such as anaemia, ulceration, haematemesis, acute pancreatitis, protein losing enteropathies, complete pyloric or small bowel obstruction, intussusception, small bowel perforation, peritonitis and even death.[16]

The diagnosis is clinical, confirmed by radiological evidence and endoscopy.

Management depends on the composition of the concretion, and includes endoscopic therapy with fragmentation, medical treatment by chemical/enzymatic dissolution and surgery.[15] Currently, various dissolving agents have been used, including cellulose, acetylcysteine, papain and coca cola.[17–20] In one study, coca cola alone achieved complete bezoar dissolution in 23.5% of patients.[17] Surgical removal should be considered in patients who fail medical therapy or who have complications such as obstruction and significant bleeding. Initial surgical therapy has been recommended in bezoars composed of vinyl gloves.[8]

Our patient had many risk factors namely fragile X syndrome, mental retardation, congenital glaucoma, institutionalisation (early deprivation) and pica disorder. In this case, a history of diaper ingestion was identified. Diaper is composed of undigested materials, such as silica crystals, which can result in the formation of bezoars.

This is a rare case report of a diaper bezoar formation and oesophagus impactation leading to a food aspiration syndrome with sudden respiratory and cardiocirculatory failure owing to an extrinsic left atrium compression.

Endoscopic techniques' management for bezoar in children are cost effective; in this case, only partial removal was achieved owing to the lack of bezoar consistency.

Medical and behaviour therapy in a long-term basis is often necessary to prevent recurrence.[2] A good prognosis is possible if psychiatric therapy is successful.

Figure 4 Upper endoscopy demonstrating the presence of silica crystals in the oesophagus.

Learning points

- Sudden stridor should be recognised as an emergency, as should be aspiration of foreign bodies in children with developmental delay.
- Pica should be recognised, especially in the institutionalised individuals or those with developmental disabilities.
- Oesophageal occlusion and foreign body aspiration to the lung may be caused by oesophageal bezoars.
- Oesophageal distension by bezoars may cause compression of the heart with resulting haemodynamic instability.
- Management of bezoar depends on the composition of the bezoar and should include not only its removal but also its prevention. Prognosis depends on the correct management of pica disorder.

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Competing interests None.

Patient consent Obtained.

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REFERENCES

1 Kay M, Wyllie R. Pediatric foreign bodies and their management. *Curr Gastroenterol Rep* 2005;7:212–18.
2 Williams DE, McAdam D. Assessment, behavioral treatment, and prevention of PICA: clinical guidelines and recommendations for practitioners. *Res Dev Disabil* 2012;33:2050–7.
3 Reddy AN. Sucralfate gastric bezoar. *Am J Gastroenterol* 1986;81:149–50.
4 Narayanan SK, Akbar Sherif VS, Babu OR, *et al*. Intestinal obstruction secondary to a colonic lithobezoar. *J Pediatr Surg* 2008;43:e9–10.
5 Evans JR, Mitchell RD. Tissue paper bezoar associated with percutaneously placed gastrostomy tube. *Gastrointest Endosc* 1988;34:142–4.
6 Perttala Y, Peltokallio P, Leiviska T, *et al*. Yeast bezoar formation following gastric surgery. *Am J Roentgenol Radium Ther Nucl Med* 1975;125:365–73.
7 Visvanathan R. Cement bezoars of the stomach. *Br J Surg* 1986;73:381–2.
8 Stringel G, Parker M, McCoy E. Vinyl glove ingestion in children: a word of caution. *J Pediatr Surg* 2012;47:996–8.
9 Phillips MR, Zaheer S, Drugas GT. Gastric trichobezoar: case report and literature review. *Mayo Clin Proc* 1998;73:653–6.
10 Burton EM, Brick WG, Hall JD, *et al*. Tracheobronchial foreign body aspiration in children. *South Med J* 1996;89:195–8.
11 Eren S, Balci AE, Dikici B, *et al*. Foreign body aspiration in children: experience of 1160 cases. *Ann Trop Paediatr* 2003;23:31–7.
12 Raza ST, Mukherjee SK, Danias PG, *et al*. Hemodynamically significant extrinsic left atrial compression by gastric structures in the mediastinum. *Ann Intern Med* 1995;123:114–16.
13 Aparna PV, Austin RD, Mathew P. PICA. *Indian J Dent Res* 2012;23:426–7.
14 Danford DE, Huber AM. PICA among mentally retarded adults. *Am J Ment Defic* 1982;87:141–6.
15 Azevedo S, Lopes J, Marques A, *et al*. Successful endoscopic resolution of a large gastric bezoar in a child. *World J Gastrointest Endosc* 2011;3:129–32.

16 Kelly ME, Ferede A, Corbally M. Hairball—a case of mistaken identity. *Ir Med J* 2010;103:214–15.
17 Okamoto Y, Yamauchi M, Sugihara K, *et al*. Is coca-cola effective for dissolving phytobezoars? *Eur J Gastroenterol Hepatol* 2007;19:611–12.
18 Ertuğrul G, Coşkun M, Sevinç M, *et al*. Treatment of gastric phytobezoars with Coca-Cola given via oral route: a case report. *Int J Gen Med* 2012;5:157–61.
19 Heinz-Erian P, Klein-Franke A, Gassner I, *et al*. Disintegration of large gastric lactobezoars by N-acetylcysteine. *J Pediatr Gastroenterol Nutr* 2010;50:108–10.
20 Koulas SG, Zikos N, Charalampous C, *et al*. Management of gastrointestinal bezoars: an analysis of 23 cases. *Int Surg* 2008;93:95–8.