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Case Report

Wide Anion Gap Metabolic Acidosis Caused by Food Protein Allergy in Infants: A 2 - Case Report

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Abstract

Here, we report two infants (aged 12 and 16 months) who presented with failure to thrive. Clinically, both infants looked normal and neither had respiratory dyspnea. However, wide anion gap metabolic acidosis was observed in the sera of both children, and both had acidic urinary pH. Specific IgE to cow's milk protein, egg white, and egg yolk was negative in the first infant, but specific IgE to cow's milk protein was positive in the second infant. The patch test for cow's milk protein and egg yolk were also positive in the second infant. Metabolic acidosis was markedly improved in both children after discontinuation of cow's milk and egg. Metabolic acidosis recurred in the first infant after he was reintroduced to cow's milk. Both children thrived and had no metabolic acidosis after dairy product and egg restriction. Both children could tolerate cow's milk and egg after one year of food restriction.

Keywords: Food protein allergy; Infant; Loss of appetite; Metabolic acidosis; Wide anion gap

Introduction

Metabolic acidosis is a clinical disturbance that is characterized by an increase in plasma acidity. Although severe episodes can be life-threatening, metabolic acidosis is sometimes a mild condition that can only be detected by blood sampling. Various conditions can be the cause of metabolic acidosis, including ketoacidosis, lactic acidosis, renal tubular acidosis and hyperchloremic acidosis. The serum anion gap has been used to identify errors in the measurement of electrolytes, to detect paraproteins, and, to evaluate patients with suspected acid-base disorders [1]. Here, we report 2 infant-aged children who presented with failure to thrive, and both had blood investigation that revealed wide anion gap metabolic acidosis.

Case Report

Case 1

A 12-month-old female infant was referred to the hospital due to a failure to gain weight for 3 months. He was diagnosed with transient renal tubular acidosis at six months of age and he has taken Shohl's solution since that diagnosis. Her parents then observed a marked decrease in his appetite during the 3-month period prior to his visit to the hospital. His past history was unremarkable. On physical examination, his weight and length were 9000g and 73 cm, respectively. Vital signs were temperature 36.5°C, respiratory rate 28min, blood pressure 10070 mmHg and heart rate 92 min. He was active, fully conscious, not distressed, and had no pallor or jaundice. Other examinations and complete blood count were unremarkable. Urinalysis revealed a pH of 5, with all of the other values being normal. His first serum electrolyte investigation showed sodium 136 mmolL, potassium 4.2 mmolL, chloride 102 mmolL and bicarbonate15 mmolL. The anion gap was 19. Blood test for specific IgE to cow's milk, soy protein, egg white, egg yolk and wheat were all negative. Dairy products and egg were then eliminated from his diet. Four weeks later, his appetite returned to normal, and his serum electrolytes were sodium 139 mmolL, potassium 4.5 mmolL, chloride 103 mmolL and bicarbonate 25 mmolL. The anion gap was 11. The Shohl's solution was then discontinued and his subsequent serum electrolyte levels remained normal. Three months later, he was challenged with cow's milk and his serum bicarbonate dropped to 15 mmolL. Acidosis was improved by dietary restriction of dairy products. After 5 months of dietary restriction, his parents reintroduced whole egg into his diet, which caused generalized urticaria. After 1 year of food restriction, he could tolerate dairy products and egg without metabolic acidosis.

Case 2

A 16-month-old, male infant was referred to the hospital due to a failure to gain weight for 4 months. He had otherwise been thriving well since birth. Since weaning, he has been regularly consuming regular baby foods and whole cow's milk daily. On physical examination, his weight and length were 10 kg and 79 cm, respectively. His vital signs were temperature 36.8°C; respiratory rate 30/min, blood pressure 9060 mmHg, and heart rate 90min. He was active, alert and had no pallor or jaundice. His first complete blood count showed eosinophil count of 1,200/mm^3. Urinary analysis showed a pH of 5, with normal results for all other values. Serum electrolytes were sodium 140 mmol/L, potassium4.6 mmol/L, chloride 104 mmol/L and bicarbonate 17 mmol/L. The anion gap was 19. Specific IgE was positive to cow's milk but negative to egg. Patch tests for cow's milk and egg yolk were positive. After elimination of dairy products and egg from his diet for 1 month, his serum electrolytes were sodium 140 mmol/L, potassium 4.6 mmol/L, chloride 104 mmol/L, and bicarbonate 22 mmol/L. The anion gap was 14. His appetite returned to normal and he increased bodyweight to 11 kg within 6 months. After one year of food restriction, dairy products and egg were reintroduced into his diet after which there was no recurrence of metabolic acidosis.

Discussion

The two cases described in the report were almost identical.

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Both children were thriving until more solid foods were gradually introduced into their diets. Except for loss of appetite, no other clinical findings of metabolic acidosis were observed in either patient. According to our review of the literature, only two other infants have been reported with metabolic acidosis caused by food protein allergy [2,3]. These patients are often misdiagnosed as renal tubular acidosis, and Shohl's solution is prescribed to treat acidosis. Although Shohl's solution could increase the serum bicarbonate in our patients, those levels were still below normal. The poor appetite that our patients with did not improve until after the correct diagnosis was made, diet restrictions were imposed, and their metabolic acidosis subsided.

We considered it is less likely that acidosis was caused by loss of bicarbonate in the urine since their urine pH levels were within acidic range. Moreover, loss of bicarbonate via the gastrointestinal tract was not considered likely since neither patient had diarrhea. Both of the immediately aforementioned conditions are almost always associated with normal anion gaps, which was not the case in our patients. The wide anion gap metabolic acidosis in our patients suggests the presence of organic acids in the blood circulation. Masson et al [2] and Rizk et al [3] reported lactic acidosis in infants with cow's milk protein intolerance. The plausible explanation for this condition is allergic reactions to foods in the small bowel, which causes mal absorption of dietary carbohydrates. The residual carbohydrate is then fermented by the bacteria in the large bowel, which results in the formation of lactic acid in the stool [4,5]. That lactic acid is then absorbed into the circulation and causes wide anion gap metabolic acidosis. The most reliable method for diagnosing food protein intolerance is the withdrawal of suspected foods, with subsequent observation for improvement in symptoms. Both of our patients showed marked improvement in metabolic acidosis within 2 weeks after withdrawal of dairy products and egg, which were both high-suspicion foods. In addition to normalization of their serum bicarbonate levels, their appetites also returned to normal. Both children began to gain weight after restriction of the suspected foods. After 1 year of avoidance of dairy products and egg, both foods were successfully reintroduced into the diets of both children with no observed reemergence of metabolic acidosis.

Clinical manifestations that include failure to thrive and loss of appetite should increase suspicion for the presence of wide anion gap metabolic acidosis caused by food protein allergy. Both children in this report returned to normal after diagnosis and restriction of suspected allergenic foods. After 1 year, both children were able to tolerate all restricted and non-restricted foods without recurrence of metabolic acidosis.

What is already known on this topic?

Until now, there is no available report on the mechanism of the 'wide anion gap' metabolic acidosis in food allergy. There were reports of severe lactic acidosis in cow's milk protein intolerance.

What this study adds?

The 'wide anion gap metabolic acidosis' is an unique manifestation of this disease which has not been reported before.

Acknowledgement

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