CASE REPORT

Anxious Hyperventilation and Hyperreflexia: Hypophosphatemia Diagnostic Clues

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Abstract

A 10-year-old boy was admitted with headache, transitory muscle spasm of the extremities, mental confusion, and shortness of breath after a stressful event. Blood tests revealed hypophosphatemia (serum phosphate 1.4 mg/dL). The diagnosis of acute hypophosphatemia due to hyperventilation, as a result of the redistribution of serum phosphate, was established. During hospitalization, he received oral phosphate supplementation and attained normal levels of serum phosphate. Further investigation revealed 25-hydroxyvitamin D deficiency and, therefore, supplementation was administered. He experienced no further similar episodes. The authors present this case to alert to the possibility of the occurrence of hypophosphatemia caused by hyperventilation in a child with a previous vitamin D deficiency.

Keywords: Anxiety/complications; Child; Hyperventilation/complications; Hypophosphatemia/diagnosis; Hypophosphatemia/drug therapy; Hypophosphatemia/etiology; Vitamin D Deficiency/complications

Introduction

Phosphate is the most abundant intracellular anion and is essential for producing adenosine triphosphate (ATP). Approximately 85% is stored in the bone as hydroxyapatite and 15% relies on soft tissue. Only 0.1% circulates in the blood stream, being free to be measured.[1] Phosphorus homeostasis is regulated by many hormones, including parathyroid hormone (PTH), and vitamin D. Hypophosphatemia is defined as a low serum phosphate concentration and classified into mild (2-2.5 mg/dL), moderate (1-2 mg/dL), and severe (< 1 mg/dL).[1] Causes of hypophosphatemia fall into three categories[2]:
- Impaired intake/absorption;
- Increased urinary excretion;
- Redistribution.

Hyperventilation fits in the third category as low serum phosphate is caused by the movement of free phosphate through cell membranes.[2]

Case Report

A Caucasian 10-year-old boy, with a three-year history of headache (normal cranio-encephalic computerized tomography) and an anxiety disorder, was admitted to the pediatric emergency department with headache, mental confusion, transitory muscle spasm of the extremities, and shortness of breath, starting in the setting of an adverse life event. Symptoms reverted spontaneously. A physical examination showed hyperreflexia. Blood analysis results were normal, with the exception of ionogram that revealed calcium 9.9 mg/dL, ionized calcium (Ca^{2+}) 1.15 mmol/L, phosphorus 1.4 mg/dL, and magnesium 1.9 mg/dL. He was hospitalized for clinical surveillance and received oral phosphorus supplementation with the normalization of the plasma levels (4.1 mg/dL). Investigation of phosphorous and calcium metabolism revealed PTH 8.8 ng/L, 1,25-dihydroxyvitamin D 46 pmol/L, and 25-hydroxyvitamin D 21 µg/L (deficit < 32 µg/L). He initiated vitamin D supplementation. Since then, he has been asymptomatic, with no more similar episodes.

Discussion

Hyperventilation decreases carbon dioxide in plasma, intracellular carbon dioxide diffuses across the cell membrane and causes the intracellular pH to rise. This intracellular rise in pH stimulates glycolysis, which requires phosphate (to make adenosine triphosphate, the major source of cell energy), and as intracellular phosphate needs increase, serum phosphate levels

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decrease.[2] Clinical manifestations of hypophosphatemia depend on the chronicity and severity of the plasma phosphate depletion. Most symptomatic patients have phosphate levels below 1 mg/dL.[1] Weakness of skeletal or smooth muscle is the most common clinical manifestation. Symptoms include tetany, myalgia, fasciculation, depressed diaphragmatic contractility and, ultimately, acute respiratory failure. Phosphate deficiency can also impair the neurologic function which may manifest as irritability, paresthesia, altered mental status, seizures and coma.[3] Although, in most cases, correcting the precipitating factors is sufficient to resolve hypophosphatemia, oral supplementation is recommended in patients with symptoms or plasma phosphate between 1 mg/dL and 2 mg/dL.[1]

In the case reported, there was a combination of both musculoskeletal and neurologic symptoms: transitory muscle spasm, transitory mental confusion, and shortness of breath. Although symptoms reverted spontaneously, the presence and maintenance of hyperreflexia on physical examination is a clue for studying phosphor-calcium metabolism. Moderate hypophosphatemia and vitamin D deficiency were found. Since phosphate and vitamin D metabolism are highly interconnected,[4] because vitamin D stimulates phosphorus gut absorption and osteoclastic bone reabsorption,[3] vitamin D deficiency may have predisposed the patient to acute hypophosphatemia in the setting of hyperventilation. Oral phosphate replacement proved effective in preventing acute recurrence. Vitamin D supplementation, by treating deficiency and restoring the body's homeostasis,[5] along with better coping strategies, decreased, respectively, the predisposition for new episodes of hypophosphatemia and the incidence of anxiety attacks.

WHAT THIS CASE REPORT ADDS

- Alert to hypophosphatemia occurrence in the setting of the hyperventilation of anxious origin.
- To emphasize the need of a meticulous physical examination to detect hypophosphatemia clinical manifestations.
- The importance of treating predisposing factors.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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Hiperreflexia e Hiperventilação Ansiosa: Pistas para Diagnóstico de Hipofosfatemia

Resumo:

Uma criança de 10 anos, género masculino, foi admitida no serviço de urgência pediátrica por cefaleia, espasmos transitórios das extremidades, confusão mental e dispneia, na sequência de um evento adverso de vida. A avaliação laboratorial inicial revelou hipofosfatemia (fosfato sérico 1,4 mg/dL). Foi feito o diagnóstico de hipofosfatemia aguda secundária a hiperventilação, por redistribuição do fosfato sérico. Durante o internamento, foi feita suplementação oral de fosfato, com normalização dos níveis séricos. O estudo alargado revelou défice de 25-hidroxivitamina D, tendo realizado, também, suplementação. Sem novos episódios semelhantes. Os autores apresentam este caso com vista a alertar para a possibilidade deste diagnóstico no contexto de uma deficiência de vitamina D.

Palavras-Chave: Ansiedade/complicações; Criança; Deficiência de Vitamina D/complicações; Hiperventilação/complicações; Hipofosfatemia/diagnóstico; Hipofosfatemia/etiologia; Hipofosfatemia/tratamento farmacológico

CASE REPORT

Hemorrhagic Shock: A Meckel Diverticulum Rare Presentation

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Abstract

Meckel diverticulum is the most frequent congenital malformation of the gastrointestinal tract. The diagnosis is challenging and often incidental as it is commonly asymptomatic. Symptomatic presentation occurs more frequently during the first decade of life and complications are more prevalent in the first two years, decreasing thereafter. Complications include ulceration with hemorrhage, perforation, bowel occlusion, and neoplasm. The authors report on a case of hemorrhagic shock as a rare presentation of a Meckel diverticulum in adolescence, along with a brief literature review.

Keywords: Adolescent; Meckel Diverticulum/complications; Meckel Diverticulum/diagnosis; Meckel Diverticulum/surgery; Shock, Hemorrhagic/etiology

Introduction

Meckel diverticulum is the most frequent congenital malformation of the gastrointestinal tract, with a prevalence of 2%-4%.[1] It is a true diverticulum containing all three layers of the intestinal wall, and originates from the incomplete involution of the omphalomesenteric duct, which establishes the communication between the yolk sac and the primitive intestine, up to the fifth to seventh weeks of fetal life.[1,2] As this obliteration fails, several malformations can occur, such as residual fibrous cord, umbilical sinus, omphalomesenteric cyst, omphalomesenteric fistula or, more commonly, Meckel diverticulum. It is located on the antimesenteric border of the ileum, approx. 46-91 cm from the ileocecal valve.[3] It may contain inclusions of heterotopic mucosa, particularly gastric (20%-57%), but also duodenal, colic, pancreatic, Brunner glands, hepatobiliary, or endometrial tissue.[1,2]

The diagnosis is frequently incidental but, when it becomes symptomatic (more often in the first decade), it may present with abdominal pain, intestinal obstruction, and gastrointestinal bleeding. About 2%-4% of patients present with complications,[2] typically before the age of 2 years, decreasing subsequently.[1] Gastrointestinal bleeding is generally related to the presence of ectopic gastric mucosa, and painless rectal hemorrhage is the most frequent presentation in children. A massive intestinal hemorrhage secondary to this congenital malformation is a rare event in the pediatric population.[4] The authors present the case of an adolescent with a Meckel diverticulum complicated with hemorrhagic shock due to massive lower gastrointestinal bleeding, and a brief review of the literature is performed.

Case Report

A 16-year-old male with a background of intussusception at 2 years of age with spontaneous resolution presented at the emergency department with 36 hours of hematochezia, vomiting, and several episodes of collpase. On physical examination, he presented a Glasgow Coma Score (GCS) of 13, with no further abnormalities on neurological examination. He was pale, tachycardiac, and hypotensive (blood pressure of 86/44 mmHg), and with a capillary reperfusion time of 4s. The abdomen was prominent with audible bowel sounds, painless, without palpable masses, or peritoneal reaction. No other abnormalities were found in the digital rectal test apart from digested blood. A bolus of 20 mL/kg of saline was given. A nasogastric tube was inserted, and no bleeding content was aspirated.

The blood tests revealed normocytic normochromic anemia with hemoglobin of 8.7 g/dL, leukocytosis of 17,470 cells/µL with neutrophilic predominance, a negative C-reactive protein, and a metabolic acidosis with lactates of 41 mg/dL.

Full hemodynamic stabilization was observed after two packed red blood cells transfusions and bleeding was controlled with an infusion of pantoprazole (80 mg followed by 40 mg every 12 hours).

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An urgent abdominal computed tomography (CT) revealed the thickening and enhancement of the wall of a diverticulum and densification of the surrounding fat, suggestive of Meckel diverticulitis (Fig. 1), without signs of pneumoperitoneum or active bleeding. In this context, a technetium-99m pertechnetate scintigraphy was made, which confirmed the diagnosis (Fig. 2).

The patient underwent a laparoscopy that identified a Meckel diverticulum with a fibrous extension to the anterior abdominal wall, and a video-assisted segmental enterectomy involving the diverticulum was performed (Fig. 3).

The postoperative period was uneventful, and he was discharged on the sixth postoperative day, clinically well, and remaining asymptomatic at two years of follow-up. The histopathology of the specimen confirmed a Meckel diverticulum with heterotopic gastric mucosa, ulcerated appearance, and free resection margins.

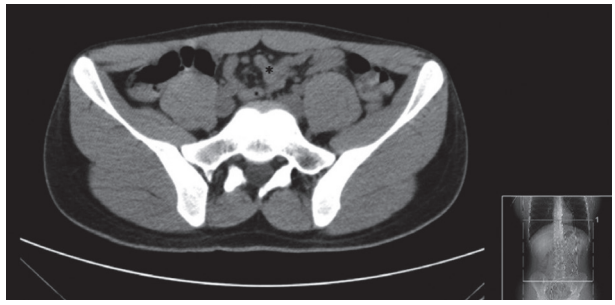

Figure 1. Abdominal computed tomography with an image compatible with Meckel diverticulum (marked by *) with thickening and enhancement of the wall of the diverticulum and densification of the surrounding fat.

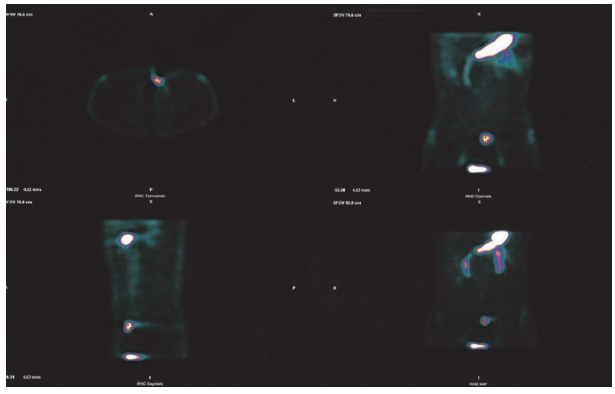

Figure 2. Scintigraphy with technetium-99m pertechnetate showing a 2 cm focal hyperfixation in the hypogastric area, left paramedian region. Fixation of the marker in ectopic gastric mucosa is compatiblVe with Meckel diverticulum.

Discussion

Lower gastrointestinal bleeding is common in the pediatric population but usually as a transiently self-limited hemorrhage. The clinical case reported illustrates a rare event, as indicated in a review of rectal bleeding causes, where only 4.2% presented with life-threatening events and, of these, only one secondary to a Meckel diverticulum.[5]

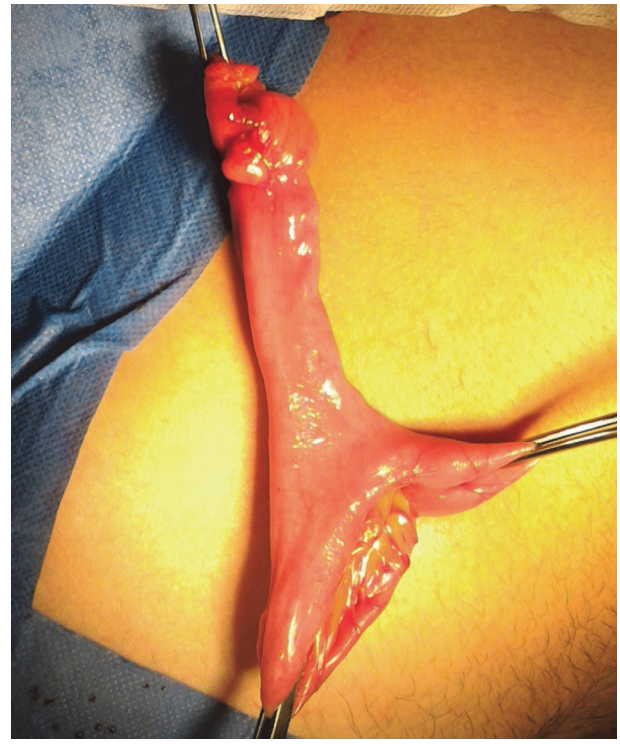

Figure 3. Surgical specimen image showing the segmental enterectomy involving the diverticulum.

The investigation of a lower gastrointestinal hemorrhage in a pediatric patient should be guided by the child's age. Symptomatic presentations of Meckel's diverticulum more commonly occur within the first decade of life. Older patients present more frequently with gastrointestinal bleeding, and younger children with obstructive symptoms.[6,7] When intussusception is the cause of obstruction, contrary to the one reported by our patient, it tends to be recurrent and atypical.

The rarity of this case is based on the complication described, which is uncommon in the patient age range, and when it occurs, it is usually less severe. According to the literature, males present a three to fourfold increased risk for complications.[1]

The diagnosis may be challenging even in symptomatic cases. A previous study reported a correct diagnosis in 88% of hemorrhagic presentations *versus* 11% in non-hemorrhagic ones.[8] In doubtful situations, in a hemodynamically unstable patient, an emergent exploratory laparotomy has to be performed.[4] In contrast, if hemodynamically stable, the exam of choice is a technetium-99m pertechnetate scintigraphy (which is a gastric mucosa tracer) that has a diagnostic accuracy of 90% in children.[9] Alternative imaging methods are the mesenteric arteriography (usually negative if bleeding < 0.5 mL/min), angio-CT which detects hemorrhages up to 0.3 mL/min, but has the disadvantage of using ionizing radiation, enterography

by magnetic resonance, endoscopic capsule, and double balloon enteroscopy.[1,10-13]

The treatment of Meckel diverticulum is guided by the clinical picture. There is considerable controversy in the literature regarding the indications for the excision of an asymptomatic, incidentally found diverticulum,[1,2] and there are no available international guidelines. On the other hand, surgical resection is the definitive treatment for a symptomatic Meckel diverticulum.

The initial management of an unstable patient involves intravenous fluid resuscitation and packed red blood cells transfusion, when clinically justifiable. In patients with gastrointestinal bleeding, proton pump inhibitors should be started, without the compromise of the scintigraphy result.[14] In the case presented, these measures contributed to the hemodynamic stabilization of the patient, avoiding an emergent surgery.

Surgical resection by laparotomy, laparoscopy, or video-assisted technique has similar satisfactory results.[4,15] Either diverticulectomy or segmental enterectomy involving the Meckel diverticulum can be made, the latter indicated whenever there is a risk of intestinal stenosis, in the presence of ischemia or perforation, if palpable changes are found in the base of the diverticulum or when the base of the diverticulum is larger than 2 cm.[1,16] In the presence of a gastrointestinal hemorrhage, segmental enterectomy is theoretically safer by resecting any possible ileal ulcers bordering the diverticulum base, and this was the procedure chosen for our patient.

The morbidity of the resection of a symptomatic diverticulum (up to 33%) is higher than in asymptomatic cases (0%-6%).[1] Mortality is rare, with an estimated incidence of 0.001%.[17] In this regard, cases such as the one presented, which may be life-threatening, are becoming less frequent.

Meckel diverticulum is an uncommon finding in adolescence and even rarer is a massive hemorrhage associated with shock as clinical presentation. The gold-standard treatment for symptomatic patients is the surgical resection of the lesion and marginal intestinal segments. In incidental asymptomatic diagnoses, surgery indication remains controversial.

WHAT THIS CASE REPORT ADDS

- Gastrointestinal bleeding can be the presentation of the Meckel diverticulum but hemorrhagic shock is rare.
- Gastrointestinal bleeding occurs generally due to the presence of ectopic gastric mucosa at the Meckel diverticulum.

Conflicts of Interest

The authors declare that there were no conflicts of interest in conducting this work.

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Consent for publication was obtained.

Confidentiality of data

The authors declare that they have followed the protocols of their work centre on the publication of patient data.

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Choque Hemorrágico: Uma apresentação Rara de Divertículo de Meckel

Resumo

O divertículo de Meckel é a malformação congénita mais frequente do trato gastrointestinal. O seu diagnóstico é desafiante e frequentemente incidental, sendo geralmente assintomático. A apresentação sintomática é mais frequente na primeira década de vida e as complicações são mais prevalentes nos primeiros dois anos, decrescendo posteriormente. As complicações incluem ulceração com hemorragia, perfuração, oclusão intestinal e neoplasia. Os autores relatam um caso clínico de choque hemorrágico como forma rara de apresentação de um divertículo de Meckel, num adolescente e fazem uma breve revisão da literatura.

Palavras-Chave: Adolescente; Choque Hemorrágico/etiologia; Divertículo Ileal/diagnóstico; Divertículo Ileal/cirurgia; Divertículo Ileal/complicações