



Letter

Food protein-induced enterocolitis syndrome triggered by orange juice

Food protein–induced enterocolitis syndrome (FPIES) is a non–immunoglobulin E (IgE)–mediated gastrointestinal disorder triggered by ingestion of certain foods. Often a delay in diagnosis occurs because patients have nonspecific symptoms and can be mismanaged as acute gastrointestinal illness, intussusception, or sepsis.¹ The most common offending foods include cow's milk, soy, and rice; however, other foods have been identified as causative agents.² Review of the literature revealed 1 case report of fruit-induced FPIES diagnosed by open food challenge (OFC) to mixed fruit including apple, pear, and banana,³ as well as 3 documented cases of FPIES to banana.^{4,5} We describe the first report to our knowledge of FPIES to orange juice.

Recently we managed a 2-year-old white boy with repetitive emesis, dehydration, and lethargy occurring on 5 separate occasions. His first reaction occurred at 10 months of age with approximately 10 episodes of emesis with no fever, diarrhea, rash, angioedema, or respiratory symptoms. The boy was admitted to our hospital and returned to baseline shortly after intravenous fluid replacement. One week later, he developed multiple episodes of emesis and was treated in the emergency department for viral gastroenteritis. At approximately 11 months of age, he had 2 similar episodes, with resolution of symptoms after intravenous fluid replacement. His last reaction, occurring at 18 months of age, was more severe, with emesis, decreased urine output, hypotension, and lethargy. He was admitted to our hospital for dehydration with negative abdominal x-ray and stool culture. Because of concern for IgE-mediated food allergy, he was referred to our allergy clinic.

In our clinic, on further history taking, the possibility of orange juice as a trigger for his reactions was identified. Emesis had occurred 1 to 2 hours after ingestion of orange juice in most of his reactions. No other specific foods or triggers could be elicited. His parents denied exposure to other citrus foods in the past. He was tolerating other fruits, including apples, pears, and bananas, thereby decreasing the likelihood of fructose intolerance. No history of atopy, including asthma, allergic rhinitis, eczema, or food allergy, was identified. The possibility of IgE-mediated food allergy was ruled out based on negative skin prick testing to orange extract and fresh orange juice. Additionally, specific IgE to orange was less than 0.1 kU/L. Given that most gastrointestinal food hypersensitivities are non–IgE-mediated and require elimination diets or food challenges for diagnosis,⁶ we elected to perform an oral food challenge (OFC). We followed the OFC protocol for FPIES outlined in “Work group report: oral food challenge testing,” by Nowak-Węgrzyn et al.⁷ Based on his history of severe reactions, we used a total challenge dose of 0.06 g protein/kg divided into 3

feedings over a total of 45 minutes with a 4-hour observation time. Consequently, he received 10 mL orange juice every 15 to 20 minutes, with a total of 30 mL given. Before the challenge, we secured intravenous access, and a complete blood count with differential was obtained. One hour and 30 minutes after the final dose, he developed repetitive emesis every 10 to 15 minutes lasting 2 hours. No urticaria, rhinorrhea, nasal congestion, wheezing, diarrhea, or other symptoms of anaphylaxis occurred during the challenge. Complete blood count with differential obtained 30 minutes into his reaction revealed a significant increase in his absolute neutrophil counts from 1,920 cells/ μ L to 3,420 cells/ μ L. Evaluation of fecal leukocytes and eosinophils was inadvertently not done. After his episodes of emesis, he became tachycardic and lethargic, for which a normal saline bolus was given. After rehydration, the boy returned to baseline with no additional management needed. Based on the positive OFC, the diagnosis of FPIES was made. We recommended our patient strictly avoid orange juice as well as other citrus products. On follow-up, our patient had experienced 1 mild episode of emesis after ingesting pineapple. He had no further emergency room visits or hospitalizations for emesis.

To our knowledge, this case is the first report of FPIES to orange juice. As is often the case with FPIES, our patient had been misdiagnosed on multiple occasions with viral gastroenteritis, and the possibility of IgE-mediated food allergy was entertained. However, based on clinical history, as well as negative skin prick testing and specific IgE level to orange juice, we opined that IgE-mediated food allergy was unlikely. We therefore proceeded to OFC and were able to confirm the diagnosis of FPIES to orange juice. When managing patients with repetitive emesis, diarrhea, dehydration, or lethargy, in addition to evaluating for common causes, we encourage clinicians to consider the possibility of FPIES induced by unusual agents such as oranges or other citrus fruits.

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