

CORRESPONDENCE

Egg yolk: An unusual trigger of food protein-induced enterocolitis syndrome

DOI:10.1111/pai.12173

To the Editor,

Recent prospective study by Katz et al. (1) reported that the cumulative incidence of food protein-induced enterocolitis syndrome is 0.34% in infants. It is characterized by profuse, repetitive vomiting, often with diarrhea that may even cause dehydration and shock after ingestion of the incriminated food (2). Because these symptoms can mimic other common pediatric conditions such as sepsis, metabolic disease, it is often misdiagnosed (3). Many foods have been implicated as triggers of FPIES, with cow's milk, soy, and grains being the commonest triggers reported. Symptoms resolve after elimination of incriminated food from the diet (4). Even though FPIES is known to be a rare disease, our observation is that the number of infants with FPIES symptoms increased in the last decade and new triggers have been reported. Previously, egg white as a causative food has been reported in a few case series or case reports, but to our knowledge there is no reported FPIES triggered by egg yolk in the literature (5–9). We aimed to emphasize that not only the whole egg and the egg white but also the egg yolk may be a trigger of FPIES in infants.

Otherwise healthy a 9-month-old boy who had experienced three episodes of repetitive and persistent vomiting after egg yolk consumption was admitted to our outpatient department. He was exclusively breastfed in the first 6 months. He ingested foods containing cow's milk such as yoghurt and cheese without any symptom. However, when he was 6 months old, he ingested a quarter of boiled egg yolk and 2 h later, he became pale and weak and had repetitive vomiting followed by diarrhea without mucus or blood. He required intravenous hydration at emergency department. At the age of 7 months, he ingested boiled egg yolk again and he experienced irritability and persistent vomiting lasting 3–4 h. Her mother reported that she separated carefully the egg yolk part from the solid egg white that was cooked completely, and she served only the egg yolk part to the baby on the previous reactions. After 3 days, he accidentally ingested a very small amount of cake containing whole egg, and similarly, he had repetitive vomiting resulting in hypotension and loss of consciousness. He recov-

ered by i.v. hydration at emergency department (the characteristics of the patient's episodes were shown in the Table 1). At the time of admission to our department, his weight and height percentiles were within normal ranges. His physical examination was normal. Complete blood count, liver, and renal function tests were normal. Serum albumin level was 4.19 g/dl. Levels of amino acids in serum and urine for metabolic diseases screening were in normal limits. Total IgE level was 4.33 kU/l. Skin prick test reactivity and food-specific IgE to egg white and egg yolk were negative. Egg yolk was eliminated in his and his mother's diet, and then, the episode did not occur. At the age of 12 months, an open food challenge test (OFC) with boiled hen's egg yolk (which consisted of 0.15 g of egg yolk protein per kilogram of body weight) was performed in our outpatient clinic. Repetitive and persistent vomiting (five times), irritability, weakness, and paleness were observed after 3 h of challenge. Even though his blood pressure was normal, he was rehydrated with saline intravenously to prevent dehydration. His symptoms resolved within 4 h. He was observed closely for 12 h in hospital. The day after, he had watery diarrhea without mucus or blood. But he had no signs of dehydration and diarrhea did not repeat.

Up to the present, there have been few case reports regarding FPIES with egg white or whole egg. In a large cohort of 161 patients, egg was the culprit food in only three of the patients (5). But recently, Hsu et al. (6) reported FPIES triggered with egg in four cases in their cohort of 38 patients in which three of four had symptoms induced by whole egg in their history. Ruffner's study, a retrospective study, is based on electronic medical records. Egg was found to be 5th most common incriminated food of FPIES and responsible for 11% of children (9). These recent studies strengthen the place of egg as a cause of FPIES.

The diagnosis of FPIES is based on clinical symptoms and exclusion of other reasons of these symptoms or positive results of OFC with causative food (2). Open food challenge test is usually suggested for patients older than 12 months because tolerance tends to develop at this age. In our patient, we

Table 1 Features of patient's episodes triggered by egg yolk

	Episode 1	Episode 2	Episode 3
Egg	Only boiled egg yolk	Only boiled egg yolk	Cake containing whole egg
Age at reaction (month)	6 month 10 day	7 month	7 month 3 day
Time of symptoms' appearance after ingestion (hour)	2	2	2
Symptoms	Repetitive vomiting followed by diarrhea without mucus or blood, weakness, paleness	Repetitive and persistent vomiting (lasting 3–4 h), irritability	Repetitive vomiting, loss of consciousness, hypotension

performed OFC with egg yolk for two reasons: first the symptoms of this patient were atypical as the egg-induced FPIES cases were presumed to be caused by egg white or whole egg and secondly to document the development of tolerance at 12 months of age. Even though egg yolk was separated carefully from egg white, cross-contamination of egg white proteins on the yolk is possible with a low probability. Previously, some reports suggested that very small quantities of the incriminated food protein may trigger FPIES (10, 11).

In conclusion, to our knowledge, this is the first case of FPIES triggered by hen's egg yolk. In the last 2 yr, the number of the documented patients with egg protein-induced enterocolitis increases in the literature, and our case report emphasizes that egg yolk may also induce enterocolitis symptoms.

Conflict of interest

All authors declare that there is no conflict of interest.

Funding

None.

Ebru Arik Yilmaz; Ozlem Cavkaytar; Ozge Uysal Soyer & Cansin Sackesen

Department of Pediatric Allergy, School of Medicine, Hacettepe University, Ankara, Turkey
E-mail: csackesen@yahoo.com

References

1. Katz Y, Goldberg MR, Rajuan N, et al. The prevalence and natural course of food protein-induced enterocolitis syndrome to cow's milk: a large-scale, prospective population-based study. *J Allergy Clin Immunol* 2011; **127**: 647–53.
2. Sicherer SH, Eigenmann PA, Sampson HA. Clinical features of food protein-induced enterocolitis syndrome. *J Pediatr* 1998; **133**: 214–9.
3. Maines E, Comberiati P, Piacentini GL, et al. Lysinuric protein intolerance can be misdiagnosed as food protein-induced enterocolitis syndrome. *Pediatr Allergy Immunol* 2013; **24**: 509–10.
4. Groetch M, Nowak-Węgrzyn A. Practical approach to nutrition and dietary intervention in pediatric food allergy. *Pediatr Allergy Immunol* 2013; **24**: 212–21.
5. Caubet JC, Nowak-Węgrzyn A. Food protein-induced enterocolitis to hen's egg. *J Allergy Clin Immunol* 2011; **128**: 1386–8.
6. Hsu P, Mehr S. Egg: a frequent trigger of food protein-induced enterocolitis syndrome. *J Allergy Clin Immunol* 2013; **131**: 241–2.
7. Fogg MI, Brown-Whitehorn TA, Pawlowski NA, et al. Atopy patch test for the diagnosis of food protein-induced enterocolitis syndrome. *Pediatr Allergy Immunol* 2006; **17**: 351–5.
8. McDonald PJ, Goldblum RM, Van Sickle GJ, et al. Food protein-induced enterocolitis: altered antibody response to ingested antigen. *Pediatr Res* 1984; **18**: 751–5.
9. Ruffner MA, Ruymann K, Barni S, et al. Food protein-induced enterocolitis syndrome: insights from review of a large referral population. *J Allergy Clin Immunol Pract* 2013; **1**: 343–9.
10. Mane SK, Hollister ME, Bahna SL. Food protein-induced enterocolitis syndrome to trivial oral mucosal contact. *Eur J Pediatr* 2013; DOI 10.1007/s00431-013-2051-2. [Epub ahead of print].
11. Tan J, Campbell D, Mehr S. Food protein-induced enterocolitis syndrome in an exclusively breast-fed infant—an uncommon entity. *J Allergy Clin Immunol* 2012; **129**: 873.