

Food Protein-Induced Enterocolitis Syndrome due to Oysters Ingestion

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Food protein-induced enterocolitis syndrome (FPIES), a non-immunoglobulin E (IgE)-mediated food allergy, manifests as profuse vomiting, often accompanied by pallor and lethargy, and sometimes diarrhea and hypotension [1]. FPIES is most commonly caused by cow's milk and soy, although a wide range of solid foods have also been reported to cause FPIES in children, particularly rice, fish and egg. Diagnosis relies on responses to elimination diets with resolution of symptoms, oral food challenges (OFCs) with reappearance of symptoms, and exclusion of other causes. The disease is typical of the early years of life; cases with adult onset are reported rarely. In the only published case in which the diagnosis of FPIES was confirmed with an OFC, the food culprit was a scallop [2]. To date, only one case of mollusc FPIES in childhood has been described. In that case, a 6 year old boy, the food culprit was the short-neck clam and the FPIES was atypical (i.e., with positive specific IgE) [3]. We present the second case of mollusc FPIES in a child, the first typical case (i.e., without detectable specific IgE).

PATIENT DESCRIPTION

The patient was a 14 year old girl who at age 11 presented with repetitive vomiting and

abdominal pain that occurred approximately 4 hours following a dinner of fish, including oysters. When the patient was 12, she reported repetitive and profuse vomiting (more severe than the previous episode) and abdominal pain, that occurred 30 minutes after a meal of oysters only (two oysters). In both episodes, she felt well before the meal, did not have diarrhea and after 3–4 hours felt well again. Moreover, on both those occasions, other diners had eaten the same foods and did not experience any adverse reaction. Prior to the first episode, the girl had eaten oysters only once, when she was 4 years old, and had no adverse reaction. After the second episode, she decided to refrain from eating oysters, but continued to eat other molluscs, crustaceans and various vertebrate fish without any adverse reaction, as she had before the critical events.

When she was 14 years old we measured her total serum IgE (18 kU/L) and specific serum IgE to oysters, crab, shrimp, mussel and cod (all < 0.01 kU/L). We also performed skin prick tests (SPT) with extract of cod and prick by prick with raw shrimp, raw squid, raw clam, raw mussel and raw oyster (all negative), and took a complete blood count (5600/ml white blood cells, including 53% neutrophils). On the same day, we conducted an open OFC with raw oysters, using 26 g (amounting to two oysters) in a single dose. Ten minutes after the first dose of oyster, the girl presented mild abdominal pain that lasted 1 hour. Two hours after the ingestion of the first dose, we administered a second dose of 30 g oyster. One hour after the ingestion of the second dose, the girl vomited five times and presented abdominal pain that lasted

1.5 hours. The patient was treated with intravenous saline infusion. Five hours after beginning the OFC she felt well and we again performed a complete blood count, which revealed elevated white blood cells (11,000/ml), with neutrophilia (74%). Eight hours after beginning OFC we repeated a complete blood count, which revealed a further rise of white blood cells (15,000/ml), with neutrophilia (80%). Eosinophil count and platelet count were within normal range each time. The patient did not demonstrate any other symptoms at home after the OFC. We also performed a fecal occult blood test and eosinophil test on a fecal sample the next day, both of which were negative. We advised the patient to avoid oysters, but allowed the ingestion of other molluscs, crustaceans and fish.

COMMENT

A literature search revealed some cases of FPIES due to shellfish ingestion in adulthood [2]. Only one case was described in the pediatric age group. That case, reported by Hayashi et al. [3], was a 6 year old boy with FPIES due to short-neck clam ingestion. He had several vomiting episodes, from age 2, after eating short-neck clams. He tested negative for short-neck clam-specific serum IgE, while the SPT was positive for short-neck clam, as was an OFC with boiled short-neck clam. He had no adverse reactions after ingestion of other molluscs and/or shellfish. The case described by Hayashi et al. [3] is compatible with the diagnosis of atypical FPIES for the presence of specific IgE against a food culprit. Our case is the first description of typical (i.e.,

without routinely detectable specific IgE) FPIES due to shellfish (oyster) ingestion in childhood.

In our patient the latency of symptoms after the ingestion was variable: 4 hours during the first episode, 30 minutes during the second one, and 3 hours during the OFC. This is not easily explained, except as a variable reactivity on different occasions. The hypothesis that the ingestion of a greater quantity of food (not only oysters) slows gastric absorption and thus the onset of symptoms, as occurred in the first episode, cannot be applied to the third episode, induced by OFC.

It is interesting that almost always, and in contrast to IgE-mediated food allergy,

patients with FPIES do not exhibit adverse reactions to other foods that are potentially cross-reactive. In fact, in our case the girl can eat other molluscs and crustaceans without any adverse reaction. A similar situation was described by Fernandez et al. [2] and Hayashi et al. [3] regarding their patients.

In conclusion, our case, like that of Hayashi et al. [3], shows that shellfish, both raw and cooked, can cause FPIES, both in the typical and the atypical form, even in children. We propose that a registry of cases with FPIES be established, since a collection of a large number of cases may help explain many aspects of this still unresolved allergic disease.

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