

Case Report



An unusual case of infant seizures with anaphylaxis to wheat

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ABSTRACT

Wheat allergy is one of the commonest food allergies in childhood and it typically presents with IgE mediated reactions, including anaphylaxis. Seizures are not typically reported to be a direct manifestation of anaphylaxis, though it can occur secondary to hypoxia following significant haemodynamic compromise. We describe a case of a previously well infant, who presented with anaphylactic shock to wheat and responded well to the initial management, but subsequently developed a cluster of brief generalised tonic clonic seizures without any ongoing haemodynamic instability. The tryptase level that was performed at 4–5 hours post reaction was raised at 49.1 µg/L. Skin prick test to wheat, wheat specific IgE, the omega-5 gliadin IgE were positive. Extensive work-up was performed to look for an underlying cause of seizures and all returned negative. To our knowledge, this is the first case report describing an unusual presentation of multiple seizures in a young infant, in association with an anaphylactic episode. In the absence of any other seizure provoking factor and underlying cause, we believe the association is more likely causative than coincidental.

Keywords: Anaphylaxis; Wheat hypersensitivity; Seizure; Child

INTRODUCTION

Wheat allergy is one of the commonest food allergies in childhood, affecting 0.4%–1% of children [1]. It has also been reported to be a frequent cause of anaphylaxis [2]. Seizures are not typically reported to be a manifestation of anaphylaxis, though it can occur secondary to hypoxia following significant haemodynamic compromise [3, 4]. We describe a case of a 5-month-old girl, who presented with anaphylaxis to wheat, and subsequent cluster of brief generalised tonic clonic seizures.

CASE REPORT

Informed consent was obtained from the patient's parents and this study was approved by Institutional Review Board of the KK Women's & Children's Hospital. The patient was born full term via normal vaginal delivery, with a birth weight of 2,756 g, and normal APGAR

scores. She was well postnatally, and was developing appropriately for age. At 5.5 months old, she had her first exposure to wheat, ingesting 3 teaspoons of wheat cereal. Within 10 minutes, she developed urticaria and periorbital angioedema. She also vomited once, and became limp and drowsy. The emergency medical service was activated and she was found by the paramedics to be hypotensive and tachycardic, with a systolic blood pressure of 53 mmHg and pulse rate of 193 beats per minute (bpm). She was saturating at 95% on room air, and was noted to be drowsy, with a Glasgow Coma Scale of 12(E3V4M5). She arrived in the hospital's Emergency Department with blood pressure of 87/37 mmHg and pulse rate of 191 bpm. She was described as alert, crying, flushed, with periorbital angioedema. Her lungs were clear and the rest of examination was unremarkable. She was promptly treated as for anaphylaxis, with intramuscular adrenaline, oral prednisolone, and oral antihistamine. She was also given an intravenous normal saline fluid bolus and transferred to the high dependency unit. In the ward, her haemodynamic status remained stable, and physical examination was normal, including her consciousness and behaviour. About 1.5 hours after the initial reaction, she started developing a cluster of 6 brief generalised tonic clonic seizures, each lasting less than 30 seconds, over the next hour. She received intravenous lorazepam, and a loading dose of intravenous phenobarbitone, before the seizures were controlled. During this period, her blood pressure was normal. An urgent computed tomography of the brain did not show any focal lesion or signs of raised intracranial pressure. A full septic workup, including a lumbar puncture was performed and she was empirically started on intravenous ceftriaxone and acyclovir. The septic workup and cerebrospinal fluid (CSF) bacterial culture and viral studies were negative. An extensive metabolic workup including CSF and serum lactate, amino acid profiles, serum acylcarnitine profile, urine organic acid profile were all not suggestive of an underlying metabolic condition. Tryptase level that was performed at 4 hours post initial reaction returned raised at 49.1 µg/L, which was in keeping with her clinical presentation of anaphylaxis. She was discharged well and advised on strict avoidance of wheat. A repeat baseline tryptase level performed 6 weeks later was normal at 8.9 µg/L. Wheat specific IgE level done at 7 months old was 10.6 kU/L, and the omega-5 gliadin IgE was 16.3 kU/L. Her skin prick test at 7 months old was initially negative to wheat (wheat, 0 mm; histamine, 5.0 mm; normal saline, 0 mm) but a repeat skin prick test performed at 13 months old was positive to wheat (wheat, 3.5 mm; histamine, 7.5 mm; normal saline, 0 mm). She was also found to be sensitised to egg and peanut on skin prick test which she was advised to avoid. When reviewed in the neurology clinic, she was doing well with no further seizures and showed normal neurodevelopment. An electroencephalogram, magnetic resonance imaging of the brain, with spectroscopy study, were all normal.

DISCUSSION

Our patient presented with anaphylactic shock to wheat and responded well to the initial management. However, her subsequent presentation of seizures was unusual, especially in the absence of ongoing haemodynamic compromise, and in the absence of metabolic disturbances. A biphasic anaphylactic reaction was considered but there were no other signs of a reaction. An underlying epileptic tendency whose first manifestation of seizures was being triggered by the anaphylactic episode was a possibility. However, with the subsequent 1-year follow-up revealing normal development and no further seizures after stopping the antiepileptic medications, this has proven to be unlikely. Adverse reactions from drugs given for the management of anaphylaxis were remote possibilities, though these have not been previously reported. The doses administered were checked and appropriate.

Seizures have been reported infrequently with anaphylaxis, and most of these case reports [3, 4] have associated hypotension, likely resulting in brain hypoxia and hence causing seizures. Falsaperla et al. [5] reviewed the literature surrounding cow's milk allergy and neurological disease, and described their case of an infant with a peculiar clinical association between gastrointestinal symptoms in cow's milk allergy, and seizures with an alteration of muscular tone. The authors postulated a possible spread of peripheral inflammatory response to the central nervous system, with disruption of the blood-brain barrier by pro-inflammatory cytokines. Complex immunological mechanisms that regulate the allergic reaction may play a key role in central nervous system manifestations. One important area studied is the alteration of blood-brain barrier as a result of immune dysfunction and inflammation. The disruption of the blood-brain barrier by the activation of brain mast cells can lead to local neuronal inflammation that could constitute an epileptogenic focus [6]. Fc-epsilon receptors which were typically thought to be expressed only by mast cells and basophils, have been identified on neurons [7]. This may mean that allergic triggers may directly affect the neurons after blood-brain barrier disruption, permitting a secondary entry of immunoglobulins. For our patient, in the absence of any other seizure provoking factor and underlying cause, we believe the association is more likely causative than coincidental. To our knowledge, this is the first case report describing an unusual presentation of multiple seizures in a young infant following an anaphylactic episode without ongoing haemodynamic instability.

In conclusion, seizures can be an unusual manifestation in a child presenting with anaphylaxis and may be related to the underlying allergic response. Further studies are required to better understand the role of neuro-inflammation in allergic diseases.

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