

CASE REPORT

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Uneventful vaginal delivery using epidural anesthesia in patient with exercise induced anaphylaxis: a case report and literature review

Kanon Morimoto¹, Kenichiro Tatsumi², Yoshitsugu Chigusa¹,
Maya Komatsu¹, Moritoki Egi², Masaki Mandai¹ and Haruta Mogami¹

¹Department of Gynecology and Obstetrics, Kyoto University Hospital, Kyoto, Japan
²Department of Anesthesia, Kyoto University Hospital, Kyoto, Japan

ABSTRACT

Exercise induced anaphylaxis (EIA) is a rare and potentially life-threatening syndrome characterized by anaphylaxis provoked by exercise. Although vaginal delivery with labor pain is a physical strain for women and a possible trigger for EIA, no consensus exists on the management strategy of delivery in patients with EIA. A 28-year-old primigravida was referred to our hospital because of history of EIA, associated with pruritus, urticaria, and respiratory distress, exacerbated during physical activity. To avoid physical stress, we chose scheduled labor induction with epidural anesthesia, and administered prophylactic intravenous hydrocortisone. She delivered vaginally with no symptoms suggestive of EIA during labor. Since it is quite possible for patients with EIA to develop anaphylaxis during vaginal delivery with labor pain, epidural anesthesia and prophylactic steroid administration may be the most rational approaches for delivery in pregnant women with EIA.

Keywords: epidural anesthesia, exercise induced anaphylaxis, pregnancy, vaginal delivery

Abbreviation:

EIA: exercise induced anaphylaxis

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INTRODUCTION

Exercise induced anaphylaxis (EIA) is a rare clinical condition characterized by anaphylaxis provoked by exercise or physical activity.¹⁻³ The incidence of EIA was estimated at 0.03% in the population of 76,229 Japanese junior-high school students,⁴ and there is no difference by gender, age, or ethnicity.³ The clinical manifestations of EIA include flushing, pruritus, urticaria, cough, dyspnea, and collapse; EIA can be life-threatening if not properly treated.^{2,3} Thus, although patients with EIA are not completely prohibited from exercising, they must be careful during physical exertion.

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Corresponding Author: Yoshitsugu Chigusa, MD, PhD

Department of Gynecology and Obstetrics, Kyoto University Hospital, 54 Shogoin Kawahara-cho, Sakyo-ku, Kyoto 606-8507, Japan

Tel: +81-75-751-3269, Fax: +81-75-761-3967, E-mail: chigusa@kuhp.kyoto-u.ac.jp

Parturition, as the term “labor” clearly indicates, is a tremendous physical strain for women, and causes exhaustion. However, no consensus exists on the management strategy of delivery in patients with EIA due to limited information. Although the prevalence of EIA is estimated to be very low, the disease concept is widely acknowledged; thus, patients with EIA are likely to become more recognized. Therefore, the safe and effective management of labor in women with EIA is crucial.

Herein, we describe a case of primigravida with known EIA and successful labor management with epidural anesthesia and prophylactic intravenous hydrocortisone.

CASE REPORT

A 28-year-old primigravida was referred to our hospital at 36^{+4/7} weeks of gestation with a history of EIA. Since childhood, she has had symptoms such as pruritus, urticaria, and respiratory distress, exacerbated during physical activity. Due to these symptoms, she was occasionally transported to the emergency department, although she had never been ventilated or admitted to the intensive care unit. Based on these episodes, EIA was diagnosed by an allergist in her early teens, and regular oral administration of an antihistamine (bilastine 20 mg/day) was initiated. Since then, the frequency of these exercise-induced symptoms has become rare; however, she developed urticaria or tightness of the chest even with mild exercise such as walking, when she missed taking the drugs. She had never been administered epinephrine for the treatment of EIA before, and she did not develop EIA during this pregnancy.

The patient was allergic to kiwis, apples, peaches, and cherries. Serum allergen-specific IgE to wheat, omega-5 gliadin, latex, and Hev b 6.02 (the most common allergens in latex) were negative. Her EIA was not caused by the ingestion of a specific food. Because her pregnancy course was uneventful, referring to previous reports, we chose scheduled labor induction with epidural anesthesia to avoid physical stress and prepare for emergencies. At 39^{+3/7} weeks of gestation, we inserted 10 mg of vaginal dinoprostone for cervical ripening, and initiated oxytocin after placement of the epidural tube at 39^{+5/7} weeks of gestation. An epidural catheter was inserted between L3 and L4, and maintenance of labor analgesia consisted of 0.1% ropivacaine with fentanyl 2 µg/mL at 10 mL/h. Two boluses of 3 mL of 0.2% ropivacaine were administered after full dilatation of the cervix for pain perception. In addition, at the onset of labor, she received 100 mg hydrocortisone intravenously and then every six hours thereafter until delivery, for a total of two doses of 50 mg. On the same day, she delivered a mature female infant weighing 2972 g with Apgar scores of 8 and 9 at 1 min and 5 min, respectively. Routine oral administration of bilastine was continued during delivery. The duration of labor was 16 h 16 min, and the patient had no symptoms suggestive of EIA during labor. Her postpartum course was also favorable.

DISCUSSION

EIA is a unique physical allergy triggered by various degrees of exercise exertion, such as walking, jogging, dancing, and tennis.⁵ EIA is basically diagnosed based on clinical history and symptoms. According to the proposed diagnostic criteria, a diagnosis of EIA is made when the following two criteria are met; (1) signs and symptoms compatible with anaphylaxis that occurred during (or within an hour) of exercise, and (2) there is no alternative diagnosis to better explain the patient's symptoms.³ EIA can be subdivided into two categories: anaphylaxis caused solely by physical activity (independent of food intake) and anaphylaxis induced by exercise after the ingestion of specific foods.³ The latter is known as food-dependent exercise induced anaphylaxis

(FDEIA), and the most common foods involved are wheat, shellfish, and legumes.³ In our case, a detailed interview revealed that the patient's symptoms were unrelated to food intake. In addition, her serum allergen-specific IgE for omega-5 gliadin was negative. Wheat omega-5 gliadin is a protein component of gluten and is the most frequent trigger for FDEIA. These findings strongly suggest that her EIA was not food-dependent.

The basic management strategy for this disease is for the patients to recognize and avoid exercise that triggers anaphylaxis. Patients should also be educated and encouraged to understand anaphylaxis-related symptoms. Prescription of epinephrine autoinjectors may also be considered.

Antihistamines are widely used as prophylactic or therapeutic pharmacotherapy for EIA. Indeed, in our case as well, the symptoms appeared without oral administration of antihistamines, suggesting that they are effective to some extent. Sodium cromoglycate and montelukast have also been reported to be effective.^{6,7} Furthermore, in recent years, reports have been accumulated showing the efficacy of omalizumab, an anti-immunoglobulin E monoclonal antibody, in cases that were refractory to other therapies.⁸⁻¹⁰

Till date, the pathophysiology of EIA is unclear; little is known about the impact of labor on pregnant women with EIA. Abnormal elevations in serum histamine during symptoms of EIA is a characteristic finding.^{11,12} A promising hypothesis regarding the mechanism of EIA is that exercise-induced changes in plasma osmolality are detected by perivascular mast cells, resulting in degranulation and anaphylaxis-related symptoms.³ Another hypothesis is that exercise disturbs systemic acid-base balance and reduces cellular pH, resulting in mast cell degradation.^{2,13} If these proposed pathophysiological mechanisms are correct (even walking caused EIA in our case), labor and vaginal delivery would be more likely to trigger EIA. However, the impact of surgical interventions such as cesarean section on this disease is unknown. Therefore, obstetricians should be well prepared for delivery in women with EIA, including measures to be taken if anaphylaxis occurs. Specifically, the intravenous route should be secured to ensure adequate infusion, and vital signs, including oxygen saturation should be monitored continuously during delivery. If anaphylaxis is strongly suspected, 0.3 mg of epinephrine must be immediately administered intramuscularly. In addition, patient information and delivery management policies were shared among the multidisciplinary staff in advance so that immediate assistance from general care physicians could be obtained in case of critical condition.

To examine the optimal delivery management for women with EIA, we conducted a literature review by searching PubMed for case reports or case series written in English using the terms "exercise induced anaphylaxis" and "pregnancy." Only five cases of EIA, including a case of FDEIA, during pregnancy with a detailed clinical course was found (Table 1).¹⁴⁻¹⁸ Smith et al described for the first time the course of two pregnancies in a woman with EIA. In their report, the patient (with a long-standing history of exercise-induced periorbital edema and urticarial) developed an anaphylactoid reaction immediately after delivery. Apart from these five cases, Shadick et al summarized the natural history of 279 patients with EIA, including seven pregnant women. Although the details are unknown, a patient had exacerbated symptoms due to exercise during pregnancy, and another had pruritus and urticaria during labor.⁵ Of note, as shown in Table 1, no anaphylaxis-related symptoms occurred in the four deliveries managed with epidural analgesia and steroid prophylaxis. Based on these facts, a similar delivery management strategy was employed in our present case. In addition to epidural analgesia, we implemented prophylactic administration of steroids because, although there are no reliable data demonstrating its effectiveness, there is also no evidence to support that steroid prophylaxis is unnecessary. In terms of steroid dosing regimen, we followed exactly what was described in the two reports.^{15,17} Consequently, the favorable outcome was obtained even though the duration of labor exceeded 16 h.

Table 1 Cases of exercise induced anaphylaxis during pregnancy reported in the literature

Author	Year	Age	GA	Parity	History of EIA	Delivery mode	Induction of labor	Anesthesia	Duration of labor	Prophylactic treatment	Event
Smith ¹⁴	1985	29	41w3d	M	Yes	Vaginal	None	None	3 h 30 min	None	Anaphylactoid reaction after delivery
Gupta ¹⁵	2007	20	N.D.	P	Yes	Vaginal	None	Epidural	12 h 30 min	Hydrocortisone, initially 100 mg followed by 50 mg 6-hourly	None
O'Connor ¹⁶	2010	29	N.D.	P	Yes	Vaginal (forceps)	None	Epidural	6 h 15 min	Hydrocortisone, ranitidine, and chlorpheniramine	None
Hindmarsh ¹⁷	2016	23	38w5d	P	Yes	Vaginal (forceps)	Oxytocin	Epidural	N.D.	Hydrocortisone, initially 100 mg followed by 50 mg 6-hourly	None
Berge ¹⁸	2022	30's	N.D.	P	Yes	Vaginal (forceps)	None	Epidural	N.D.	Glucocorticoid 100 mg, and antihistamines (cetirizine) 10 mg	None
Ours	2023	28	39w5d	P	Yes	Vaginal	Oxytocin	Epidural	16 h 16 min	Hydrocortisone, initially 100 mg followed by 50 mg 6-hourly	None

GA: gestational age

EIA: exercise induced anaphylaxis

P: primigravida

M: multigravida

N.D.: not described

In conclusion, given the pathophysiology of EIA and previous reports, it is quite possible that pregnant women with EIA develop anaphylaxis with labor pain during vaginal delivery. Our case indicates that at present, epidural anesthesia and prophylactic steroid administration are the most rational approaches for delivery in pregnant women with EIA. Advance preparation of treatment for anaphylaxis is equally essential.

ETHICAL APPROVAL

Ethical approval was not required for this study in accordance with the local guidelines of Kyoto University Hospital.

INFORMED CONSENT

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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