Letter to the Editor

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Perioperative management of a patient with hereditary angioedema undergoing oral surgery

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Dear Editor:

Hereditary angioedema (HAE) is an edema of the subcutaneous or submucosal tissues caused by emotional stress, physical stress or other triggers. HAE can cause fatal upper airway obstruction, so caution is required when treating these patients [1]. We describe a case in which a patient with suspected HAE safely underwent oral surgery under general anesthesia. Written informed consent was obtained from the patient.

A 36-year-old woman (height 160 cm; weight 46 kg), underwent extraction of a horizontally impacted wisdom tooth. She had a history of angioedema and was taking tranexamic acid and lupatadine fumarate. The patient often had edema of the face, larynx, and hands at night and dawn, for which she was taking prednisolone. Considering the possibility of airway obstruction caused by perioperative angioedema, the patient was referred to the otolaryngology and dermatology departments before surgery to establish a perioperative backup plan (emergency tracheostomy). In preparation for the occurrence of edema, especially laryngeal attack, the hospital pharmacy prepared dry concentrated human C1-inactivator (Verinert® P, CSL Behring, Tokyo, Japan). Sodium hydrocortisone succinate was administered intravenously and diazepam was administered orally before the patient entered the operating room (OR). In the OR, d-chlorpheniramine maleate, famotidine, and tranexamic acid were administered intravenously. Then, propofol was administered together with remifentanil hydrochloride, fentanyl citrate, and rocuronium bromide. Next, a 7.0 mm diameter tracheal tube was inserted orally using a McGrath MAC video laryngoscope (Covidien Japan, Tokyo, Japan). Cuff pressure was adjusted to 20 cmH_2O. Anesthesia was maintained with sevoflurane, and there were no vital changes or problems during surgery. After the operation, we observed the glottis and trachea with a bronchoscope to confirm the absence of edema or secretion retention. We performed a cuff-leak test to confirm the absence of upper airway stenosis. We then administered sugammadex sodium to induce spontaneous breathing. After stable spontaneous breathing was confirmed, sevoflurane administration was stopped and the patient was extubated under deep anesthesia. After confirming that the airway was still open after extubation, and there were no other

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complications, the patient was returned to the ward. SpO_2 monitoring was continued, and no decrease in SpO_2 was observed next morning. The patient was discharged from hospital on the third postoperative day.

HAE is an autosomal dominant disorder [2] caused by mutations in the SERPING gene encoding C1 inhibitor (C1-INH) and can result in a quantitative (HAE type I) or functional (HAE type II) deficiency of C1-INH [1]. The prevalence of HAE types I and II is estimated to be approximately 1 in 50,000, and the prevalence of HAE type III is believed to be much lower, although the true prevalence is unknown [1]. Although this patient had normal C1-INH levels and function and no mutation in the SERPING1 gene, the presence of routine episodes of edema suggested the possibility of HAE type III.

Recommendations for the anesthetic management of HAE include prevention and treatment of acute attacks, but the importance of anxiety control is underestimated [3]. Rosa et al. [4] stated that careful management of anxiety in patients with HAE is important to prevent attacks. A previous review showed that patients with systemic diseases such as HAE may be at higher risk of developing acute attacks in the perioperative period than healthy subjects because they are already anxious before surgery [3]. Therefore, we decided to administer diazepam to ensure adequate anxiety control. The same review [3] reported that sedation is preferable to general anesthesia because intubation itself may induce an acute attack. However, in this case, the wisdom tooth was severely impacted, and invasive surgery and stress were anticipated. Therefore, we decided to use general anesthesia with sufficient precautions according to the guidelines [5], which emphasize the importance of multidisciplinary collaboration in the perioperative management plan for HAE. We requested a consultation with a dermatologist treating HAE patients and requested a backup plan from the otolaryngology department in the event that an emergency tracheostomy might be necessary.

We achieved safe perioperative management of a patient with suspected HAE. It is important to modulate or eliminate possible precipitating factors as much as possible, and to collaborate with other departments in case of emergency.

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