Commentary

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Uncommon Manifestations of Mitochondrial Encephalomyopathy (MELAS) and Diabetes

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Description

The syndrome of Mitochondrial Encephalomyopathy, Lactic Acidosis, and Stroke-like episodes (MELAS) is a multi-organ disease that develops mainly in childhood and is a type 1 progressive mitochondrial encephalomyosis. Its clinical features include multiple cerebral infarctions, progressive muscle weakness, peripheral neuropathy such as numbness in the hands and feet, endocrine symptoms, sensorineural hearing loss, intellectual regression, and many other symptoms due to mitochondrial genetic abnormalities. Because of the wide variety of systemic symptoms, it takes time to reach a diagnosis. There is no clear cure, and it is recognized as a disease with a poor prognosis. There are reports of a decrease in general muscle strength, but there have been a few reports of symptoms around the oromaxillofacial area. In this study, a patient with type 1 diabetes mellitus whose jaw position was unstable due to a decrease in the strength of the masticatory muscles thought to be caused by MELAS underwent systemic management in cooperation with internal medicine, tooth extraction under local anesthesia, and then all-jaw oral function management, with a good course. The volume and form of meals improved. The patient received regular oral care after dental treatment, but then refused dental treatment. After that, the patient's general condition rapidly deteriorated, and he died due to multiple organ failure. The syndrome of Mitochondrial Encephalomyopathy, Lactic Acidosis, and Stroke-like episodes (MELAS) is a maternally inherited disease caused by mutations in mitochondrial DNA. The onset has two peaks, with high frequencies in childhood and adulthood, and it presents with a wide range of systemic symptoms, so it may take time to be diagnosed. There is no specific treatment, only symptomatic treatment, and it is recognized as a disease with a poor prognosis.

Desmopressin Therapy

Oral problems in patients with MELAS have been reported in cases where oral cleaning became difficult due to muscle weakness, cases of dysphagia, and cases of worsening of the oral condition due to diabetes mellitus, but no manifestations in the stomatognathic region have been reported so far. For MELAS patients who require oral surgery, their general condition needs to be managed according to each symptom. In the present case, systemic management was performed in collaboration with internal medicine for a patient with type 1 diabetes mellitus who was unable to stabilize the jaw position due to progressive weakening of the masticatory muscles thought to be caused by MELAS. Tooth extraction was performed under local anesthesia, and all-jaw oral function management was then performed. Diabetes Insipidus (DI) is a rare clinical condition in the postoperative period. Post-surgery polyuria is a common finding, as the body excretes the excessive fluid given during surgery. It is important to diagnose and differentiate the DI from post-operative polyuria, as DI can lead to severe dehydration and electrolyte disturbances. We report two unusual cases of perioperative DI requiring desmopressin therapy. A 46-year-old healthy male patient developed intraoperative DI leading to hypernatremia during the anterior cervical discectomy and fusion. Anesthesia was maintained with protocol and remifentanil Target-Controlled Infusion (TCI). After two hours of surgery, the patient became polyuric and was passing diluted urine. He received desmopressin and hydration. The patient recovered and was transferred to the ward, then, discharged home without any clinical or neurological problems.

A 36-year-old healthy male patient underwent elective 3rd ventricular cyst excision. Pre-anesthesia assessment did not reveal any comorbidities and the surgery was uneventful. His anesthesia was maintained with propofol and remifentanil TCI (Target-Controlled Infusion). In the postoperative period, he developed DI requiring hydration and desmopressin. The patient's further recovery was uneventful. He was discharged home through the ward. A 59-year-old Japanese woman with a 22-year history of long-term hemodialysis was admitted to our hospital for further examination of hyperglycemias and anemia. Five months before hospitalization, her fasting plasma glucose value was 99 mg/dL and her glycated hemoglobin was 5.7%. On admission, her fasting plasma glucose value was 873 mg/dL, glycated hemoglobin was 16.2%, C-peptide reactivity was 22.3 ng/mL (reference range, 0.5-3.0, 22.3 ng/mL), and Homeostasis Model Assessment of Insulin Resistance (HOMA-IR) was 10.6 (reference range, <2.0); the high HOMA-IR indicated high insulin resistance. Intensive insulin therapy was started for hyperglycemia, which required more than 40 units/day. Computed tomography showed a hyper vascular lesion 2.2 cm in

diameter on the right kidney; therefore, right nephrectomy was performed. Complete resection was confirmed, and the lesion was diagnosed as a clear cell type of Renal Cell Carcinoma (RCC). Immediately after nephrectomy, glycemic control normalized and administration of insulin was discontinued. Fourteen days after nephrectomy, the HOMA-IR decreased to 2.96. RCC that develops in patients receiving long-term hemodialysis has been reported to be dialysis-related RCC, but there have been no reports suggesting a relationship between dialysis-related RCC and diabetes. To our knowledge, this is the first report of RCC presenting with the para neoplastic syndrome of acute-onset diabetes because of insulin resistance. Pancreatic cancer is rarely diagnosed during pregnancy; it usually manifests with symptoms such as epigastria pain, vomiting, weight loss, and jaundice, rarely mimicking the hemolysis, elevated liver enzymes, and low platelet count syndrome.

Several Neurosurgical Conditions

It has been postulated that there exists a correlation between the diagnosis of gestational diabetes mellitus and the occurrence of pancreatic cancer later in life. We conducted an expert literature review of the 31 available documented pancreatic cancer cases that were diagnosed during pregnancy. We also report pancreatic adenocarcinoma incidentally suspected in an asymptomatic woman affected by gestational diabetes mellitus; the woman was undergoing a fetal growth scan. Neuroendocrine dysfunction is a common complication of several neurosurgical conditions. In particular, Central Diabetes Insipidus (CDI) can occur subsequent to traumatic brain injury, subarachnoid hemorrhage, and cerebral tumors or as a result of a complication following pituitary neurosurgery. In contrast, surgical resection of non-sellar tumors does not commonly result in CDI, with only a few cases reported in the literature. We report the case of a 40-year-old man who presented a transient CDI following surgical resection of a pineal papillary tumor *via* an occipital interhemispheric trans-tentorial approach. The underlying pathogenesis of CDI occurring post resection of tumors arising at a distance from the sella is not yet clearly understood, especially since there is no evidence of direct compression of the pituitary stalk. With regards to our case, we hypothesize that restauration of the initial obstructive hydrocephalus might induce a rapid intracranial pressure variation leading to hemodynamics changes of the portal hypophysis vascular system. Postoperative air entrapment in the sellar region might also lead to irritation of the pituitary stalk.