Case Report

Hunter’s glossitis and autoimmune gastritis: a case report

N. Oda1, H. Takakura2, M. Maeda1,3 and I. Takata1

1Department of Internal Medicine, Fukuyama City Hospital, Fukuyama, Japan
2Department of Oral and Maxillofacial Surgery, Fukuyama City Hospital, Fukuyama, Japan
3Department of Kidney, Diabetes, and Endocrine Diseases, Okayama University Hospital, Okayama, Japan

Address correspondence to Dr N. Oda, Department of Internal Medicine, Fukuyama City Hospital, 5-23-1 Zao-cho, Fukuyama 721-8511, Japan. email: pgj77o3@s.okayama-u.ac.jp

Learning points for clinicians
When encountering glossitis in patients with a history of autoimmune thyroid diseases or type 1 diabetes mellitus, the possibility of vitamin B12 deficiency and autoimmune gastritis should be considered. Timely recognition and management of these conditions are crucial to prevent complications such as pernicious anaemia and neuropathy.

Case presentation
Our patient was a Japanese woman in her 50s who had developed type 1 diabetes mellitus and Hashimoto’s during childhood. These conditions had been treated with insulin and thyroid hormone replacement therapy. Her family history was remarkable for Hashimoto’s thyroiditis in her father. She also had a history of bronchial asthma. When her control over the bronchial asthma had recently deteriorated, necessitating an increase in inhaled corticosteroid medication, the patient began experiencing recurrent glossitis. Initially, this was suspected to be an adverse reaction to the inhaled corticosteroids and was managed via gargling and changing the inhalation device. However, her glossitis worsened over time and she developed anaemia as well. Her tongue exhibited severe atrophy, appeared red and smooth and was painful with a burning sensation, as well as dysgeusia (Figure 1A). Notably, no neurological symptoms were observed. Her haemoglobin levels decreased from 12.9 to 10.7 g/dl over 1 year, and she presented with macrocytic anaemia characterized by a mean corpuscular volume (MCV) of 106 fl and a mean corpuscular haemoglobin (MCH) of 35.8 pg. Her serum lactate dehydrogenase, ferritin and folic acid levels were elevated, measuring 414 U/l, 219.4 ng/ml and 14.8 ng/ml, respectively. Her vitamin B12 levels were decreased, at 118 pg/ml, but her iron levels were within the normal range, at 78 μg/dl. Gastrointestinal endoscopy revealed advanced atrophic gastritis primarily affecting the gastric corpus (Figure 1B). The patient tested positive for antiparietal antibodies but was not infected with Helicobacter pylori. These findings led to a diagnosis of Hunter’s glossitis, pernicious anaemia and autoimmune gastritis (AIG). Following treatment with vitamin B12 intramuscular injections, the glossitis quickly improved and the patient’s laboratory results normalized, with haemoglobin levels reaching 12.8 g/dl, MCV stabilizing to 93.9 fl

Figure 1. (A) Severe tongue atrophy with a smooth, reddened appearance. (B) Gastrointestinal endoscopy revealing advanced atrophic gastritis primarily affecting the gastric corpus.
and MCH falling to 31.6 pg after 2 months. Maintenance treatment with intramuscular vitamin B12 injections is ongoing.

Discussion

AIG is an autoimmune disease characterized by cellular immunity that leads to the production of anti-parietal cell antibodies and the destruction of parietal cells. This results in an acid-free environment in the gastric corpus, ultimately causing hypergastrinemia. The endoscopic hallmark of AIG is advanced atrophy predominantly affecting the gastric corpus, which distinguishes it from the vestibular-dominant atrophy observed in atrophic gastritis associated with H pylori infection. AIG also carries a high risk of developing gastric neuroendocrine tumours and gastric cancer, largely because of hypergastrinaemia and advanced gastric corpus atrophy. Reduced secretion of intrinsic factors in AIG leads to impaired vitamin B12 absorption, culminating in pernicious anaemia and subacute combined degeneration of the spinal cord in later stages. Atrophic glossitis resulting from vitamin B12 deficiency, often called Hunter’s glossitis, precedes the onset of pernicious anaemia and neuropathy. Recognizing Hunter’s glossitis is crucial to identifying vitamin B12 deficiencies.

In this case, we considered the likelihood of underlying autoimmune polyendocrine syndrome (APS), a rare condition that affects multiple endocrine glands. Specifically, APS type 3 was considered, given the coexistence of autoimmune thyroiditis and non-thyroid autoimmune conditions excluding autoimmune adenitis—namely, type 1 diabetes mellitus and pernicious anaemia. APS type 3 is frequently observed in middle-aged women and may involve a combination of genetic and environmental factors. Notably, some genes associated with AIG are located at the same loci as diabetes susceptibility genes, offering a plausible explanation for the strong association between AIG and type 1 diabetes mellitus in humans.

Although common, glossitis requires particular attention in patients with autoimmune thyroid diseases or type 1 diabetes mellitus. This should prompt a comprehensive investigation focusing on potential vitamin B12 deficiency and AIG.

Author contributions
Naohiro Oda (Conceptualization [lead], Writing—original draft [lead]), Hiroaki Takakura (Writing—review & editing [equal]), Megumi Maeda (Writing—review & editing [equal]) and Ichiro Takata (Writing—review & editing [equal])

Consent for case reports
Written informed consent was obtained from the patient.

Conflict of interest
None declared.

References