

## A rapid recovery from pagophagia following treatment for iron deficiency anemia and TMJ disorder accompanied by masked depression

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**ABSTRACT** Pagophagia, or ice eating, is a particular expression of the more general phenomenon of pica and is closely associated with the development of iron deficiency anemia (IDA). The diagnosis of pagophagia and its treatment are not a common part of oral and maxillofacial practices. This article presents the first reported case of rapid recovery from prolonged pagophagia following treatment for IDA and temporomandibular joint (TMJ) disorder accompanied by masked depression.

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**Key words :** Temporomandibular joint disorder, Pagophagia, Iron deficiency anemia, Masked depression

### CASE REPORT

A 37-year-old Japanese woman was referred to the Department of Oral Surgery, Kawasaki Medical School Hospital with a history of temporomandibular joint (TMJ) pain on the left side of about one week' duration. The patient slept well and had a good appetite without GI tract symptoms, but also complained of fatigue and a depressive mood. In early adolescence, the onset of ice eating occurred, and the patient had continued to crush large numbers of ice cubes (30 or more/day) with her left teeth for more than 20 years. However her craving for ice cubes stopped during administration of iron therapy after her second pregnancy. There was no desire to eat other nonnutritive substances, such as dirt, clay or starch. Her other medical history was myoma uteri with a little excessive menstrual blood loss. At her first examination in

our clinic, the maximal opening of the mouth was 44mm. There was TMJ pain on the right side with bilateral clicking. The tongue was not atrophic and was without pain (Fig.1). Neither xerostomia nor any taste abnormality was apparent. A panoramic radiogram disclosed a reduced joint space of the left TMJ and asymptomatic caries of the upper left



Fig. 1. View of the tongue at the first examination

third molar (Fig.2). MRIs of the left TMJ disclosed a thin, anterior displaced disk with reduction (Fig.3), but no abnormality was present in the right TMJ. Laboratory studies revealed iron deficiency anemia (IDA) with hemoglobin 10.5 g/dl (11.5-14.5), MCV 66.6 fl (87-99), serum iron 12  $\mu$ g/dl (51-203), serum ferritin 2ng/ml (5-160) and serum transferrin 325mg/dl (200-340). The self-rating questionnaire for depression (SRQ-D) was used to evaluate depressive mood. A full score for the SRQ-D is 36 points. The SRQ-D is suitable for evaluating masked depression. It includes many questions concerning depression-related physical symptoms, and a score of 12 or higher points indicates possible masked depression<sup>1)</sup>. The SRQ-D of the patient revealed a high score (24 points) suggestive of masked depression. The tentative diagnosis was coexistence

of temporomandibular joint disorder (local synovitis of the TMJ) with prolonged pagophagia and masked depression. The patient consulted a psychiatrist on the same day and started on an antidepressant (selective serotonin reuptake inhibitor: SSRI) and a benzodiazepine tranquilizer with a diagnosis of depression, obsessive-compulsive disorder and panic disorder. The patient stopped ice eating because of the TMJ pain. One week later, the TMJ pain disappeared. However a recurrence of ice eating took place immediately after resolution of the TMJ pain even with administration of SSRI and the benzodiazepine tranquilizer. There was the possibility that the TMJ pain may have originated from both prolonged pagophagia and depression. Treatment for IDA with pagophagia was initiated with oral sodium ferrous citrate, 200mg/day, to



Fig. 2. Panoramic radiogram at the first examination

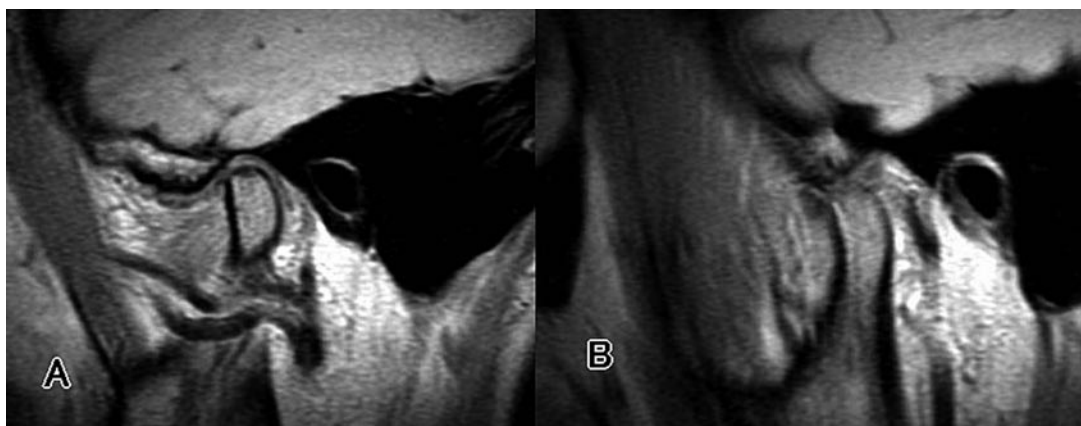


Fig. 3. MRIs of the left TMJ on a proton-density-weighted image (A) Sagittal closed-mouth image and (B) open-mouth image

be replete with iron. Her pagophagia as well as depressive mood with fatigue dramatically stopped within a few days after administration of oral sodium ferrous citrate. One month after the iron therapy, laboratory studies revealed hemoglobin 13.0 g/dl, MCV 78.6 fl, serum iron 62 µg/dl, serum ferritin 10ng/ml and serum transferrin 283 g/dl. The patient consulted a hematologist, but there were no apparent causes of blood loss except for a little excessive menstrual blood loss. Follow-up at three months revealed neither TMJ pain nor ice eating.

## DISCUSSION

Patients with iron deficiency may or may not exhibit one or a few signs and symptoms considered highly specific for iron deficiency, namely, pagophagia, koilonychia, and blue sclerae<sup>2)</sup>. The diagnosis of pagophagia and the treatment related to it are not a common part of oral and maxillofacial practices. According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR)<sup>3)</sup>, pica is “the persistent eating of nonnutritive substances for a period of at least one month, without an association with an aversion to food”. Diagnosis also requires that the eating of nonnutritive substances be inappropriate to the developmental level and “sufficiently severe to warrant independent clinical attention” secondary to anemia, bowel obstruction, or lead poisoning. The most commonly consumed substance is ice (pagophagia)<sup>4)</sup>. Pagophagia was detected in 8.1% of urban, pregnant, otherwise healthy African-American women of low socioeconomic status with low serum ferritin suggesting IDA<sup>4)</sup>. The prevalence of IDA varies widely depending on a variety of factors, such as age, ethnic composition, socioeconomic status, and dietary habits such as vegetarianism<sup>5)</sup>. The frequency of IDA was as high as 9.1% among a 10 to 19-year-old cohort of apparently healthy women in a metropolitan area of Japan<sup>6)</sup>. In the last 15 years<sup>7)</sup>, anemia

(hemoglobin <12.0g/dl) increased from 16.8% to 20.6% in Japanese women aged 30-39 years old, and from 20.2% to 26.9% in Japanese women aged 40-49 years old, but there have been no reports of pagophagia in the Japanese literature, suggesting that the incidence of pica may be extremely rare in Japan.

The main causes of iron deficiency are menstruation and GI tract blood loss, including hiatal hernia, esophageal varices, gastritis, duodenitis, peptic ulcer, cholelithiasis, intrahepatic bleeding, inflammatory bowel disease, diverticulosis, hemorrhoids, and adenomatous polyps<sup>2)</sup>. In women of childbearing age<sup>2)</sup>, genitourinary blood loss with menses is often responsible for increased iron requirements, as in the present case.

Atrophic glossitis, angulus vitiosus and Plummer-Vinson syndrome are the most well-known oral manifestations associated with IDA. Pagophagia associated with the development of IDA causes dental complications such as erosion, abrasion, loss of vertical dimension, cracked teeth, teeth sensitivity and recurrent decay<sup>8)</sup>. There were none of the above-mentioned symptoms in the present case. To our knowledge, this article presents the first reported case of pagophagia and TMJ disorder accompanied by masked depression.

The present case was also suggestive of masked depression as evaluated with the SRQ-D and the final diagnosis by a referred psychiatrist was depression, obsessive-compulsive disorder and panic disorder. SSRI is a suitable drug for these disorders and was prescribed for the present case. Coexistence of the depressive mood accompanying TMJ pain and prolonged pagophagia was resolved within a few days after iron therapy for IDA. Iron therapy can alter brain function, such as verbal learning and memory<sup>9)</sup>. However, Fordy and Benton measured ferritin concentrations and found no association between low iron status and psychological function<sup>10)</sup>. The association between iron status and pagophagia

may be complex, and further research is needed.

The goal of therapy for IDA is to supply sufficient iron to repair the hemoglobin deficit and replenish storage iron and underlying diseases if they exist. An increase in the hemoglobin concentration of at least 2 g/dl after three weeks of iron therapy for IDA is generally used as the criterion for an adequate therapeutic response and oral iron should be continued until the plasma ferritin concentration exceeds 50  $\mu\text{g/l}$  (50 ng/ml)<sup>2)</sup>. In the present case, pagophagia stopped within a few days after iron therapy, suggesting that the amount of iron sufficient for stopping pagophagia may be small.

## REFERENCES

- 1) Rockliff BW: A brief self-rating questionnaire for depression (SRQ-D). *Psychosomatics* 10: 236-243, 1969
- 2) Brittenham GM: Disorders of iron metabolism: iron deficiency and overload. In *Hematology, basic principle and practice*, third edition (Hoffman R, et al. ed). New York, USA, Churchill-Livingstone. 1999, pp410-417
- 3) American Psychiatric Association: *Diagnostic and Statistical Manual of Mental Disorders*, 4<sup>th</sup> Edition, Text Revision. Washington, DC, American Psychiatric Association, 2000
- 4) Edwards CH, Johnson AA, Knight EM, Oyemade UJ, Cole OJ, Westney OE, Jones S, Laryea H, Westney LS: Pica in an urban environment. *J Nutr* 124(Suppl): 954S-962S, 1994
- 5) Panagiotou JP, Douros K: Clinicolaboratory findings and treatment of iron-deficiency anemia in childhood. *Pediatr Hematol Oncol* 21: 521-534, 2004
- 6) Kanamaru A: Iron deficiency anemia. *Nippon Rinsho* 66: 499-504, 2008
- 7) Hayashi F, Yoshiike N, Yoshita K, Kawahara K: Trends in the prevalence of anaemia in Japanese adult women, 1989-2003. *Public Health Nutr* 11: 252-257, 2008
- 8) Johnson CD, Koh SH, Shynett B, Koh J, Johnson C: An uncommon dental presentation during pregnancy resulting from multiple eating disorders: pica and bulimia: case report. *Gen Dent* 54: 198-200, 2006
- 9) Bruner AB, Joffe A, Duggan AK, Casella JF, Brandt J: Randomised study of cognitive effects of iron supplementation in non-anaemic iron-deficient adolescent girls. *Lancet* 348: 992-996, 1996
- 10) Fordy J, Benton D: Does low iron influence psychological functioning? *J Hum Nutr Diet* 7: 127-133, 1994