

Case Report

A rare case of a tongue mass in a patient with megaloblastic anemia

Hitoshi Osano^{1,2}, Toru Miyauchi², Masaki Toshima³
Koichi Matsumoto², and Mikio Kusama²

Abstract

Megaloblastic anemia is most commonly due to a deficiency of folate or vitamin B₁₂. This report describes the clinical features of a case of megaloblastic anemia, in which an oral symptom was the first significant finding. A mass on the tongue as an initial symptom of megaloblastic anemia is rare. We report a 69-year-old male patient who had a tongue mass, fissured tongue, and various laboratory abnormalities. It is interesting that his mass reduced with the normalized serum level of vitamin B₁₂ and iron after substitution therapy.

(Key words : deficiency of vitamin B₁₂, megaloblastic anemia, tongue mass)

Introduction

Megaloblastic anemia is most commonly due to a deficiency of folate or vitamin B₁₂. Folate deficiency is usually nutritional in origin and may be seen in alcoholics and poor elderly people, but also in patients on hyperalimentation or hemodialysis. The most common cause of vitamin B₁₂ deficiency is pernicious anemia, a condition in which the portion of gastric mucosa that contains the parietal cells is destroyed through an autoimmune mechanism. Clinical symptoms in the oral mucous membranes have been reported to occur in 50% of all patients with megaloblastic anemia¹. Burning and itching sensations in the tongue, a slight irregular red enanthema of the buccal mucosa, and atrophy of the filiform and later the fungiform papillae of the tongue leading to a completely atrophic, smooth varnished fiery red surface of the tongue (Hunter's glossitis) are the most frequent changes. In early cases, usually only the tip of the tongue and the margins are slightly red and atrophic, whereas the mucous membranes of the entire oral cavity appear pale. The tongue may be fissured, but is more often lobulated. We report a case of megaloblastic anemia in which an enlarged mass of the tongue was the first significant clinical finding.

Case report

A 69-year-old male was referred to our hospital by his physician for a tongue mass. He first noticed the red-bean sized mass on his tongue about 4 months previously, and it had rapidly increased in size over several weeks before initial consultation.

The patient's previous history showed nothing of note. The patient complained of a feeling of fatigue when he took slight exercise one month previously.

1. Division of Dentistry, Oral and Maxillofacial Surgery, Department of Medicine, Saitama Medical Center, Jichi Medical University
2. Division of Dentistry, Oral and Maxillofacial Surgery, Department of Medicine, Jichi Medical University
3. Division of Hematology, Department of Medicine, Jichi Medical University

Extra-oral findings revealed no abnormalities. Intra-oral examination revealed a tender lobulated mass with pedunculated and diffuse margins on the dorsum of the tongue ; the mass had a maximum diameter of 14.0 mm, was deep-reddish in colour, with alternating paler or pinkish areas, and the tongue was fissured (Fig. 1). The lump was firm on palpation, non-fluctuant, not attached to deep tissue layers, painless, and non-bleeding. No other evidence of oral disease was noted. On the basis of the clinical appearance, a differential diagnosis was formulated ; this included fibroma, physical injury, pyogenic granuloma, and hemangioma. The aim of treatment was to excise the mass. A preoperative blood test was performed. Remarkable symptoms of anemia were detected (Table 1). Investigations showed hemoglobin at 5.4 g/dl, a total leukocyte count of 4300 cells/mm³ and a normal differential count, a platelet count of 69,000/mm³ and a serum vitamin B₁₂ level of 71.3 pg/ml.



Figure1 Initial appearance of the lesion

| | | Laboratory Findings | | | |
|---------------------|-----------------------------|----------------------------|---------------|------------------|--------------|
| 〈Blood examination〉 | | 〈Chemistry panel〉 | | 〈Serologic test〉 | |
| WBC | 4.3 × 10 ³ /μl | TP | 6.2 g/dl ↓ | Folic acid | 2.8 ng/ml |
| RBC | 129 × 10 ⁴ /μl ↓ | ALB | 3.9 g/dl | VB12 | 71.3 pg/ml ↓ |
| Hb | 5.4 g/dl ↓ | BUN | 13 mg/dl | | |
| Ht | 15.8 % ↓ | CRE | 1.01 mg/dl | | |
| MCV | 122 fl ↑ | T-BIL | 1.66 mg/dl ↑ | | |
| MCH | 41.8 pg ↑ | D-BIL | 0.13 mg/dl | | |
| MCHC | 34.1 % | GOT | 18 mU/ml | | |
| Plt | 6.9 × 10 ⁴ /μl ↓ | GPT | 10 mU/ml | | |
| Reticulocyte | 15 % | LDH | 1,113 mU/ml ↑ | | |
| | | ALP | 274 mU/ml | | |
| | | γ -GT | 14 mU/ml | | |

Table1 First clinical examination

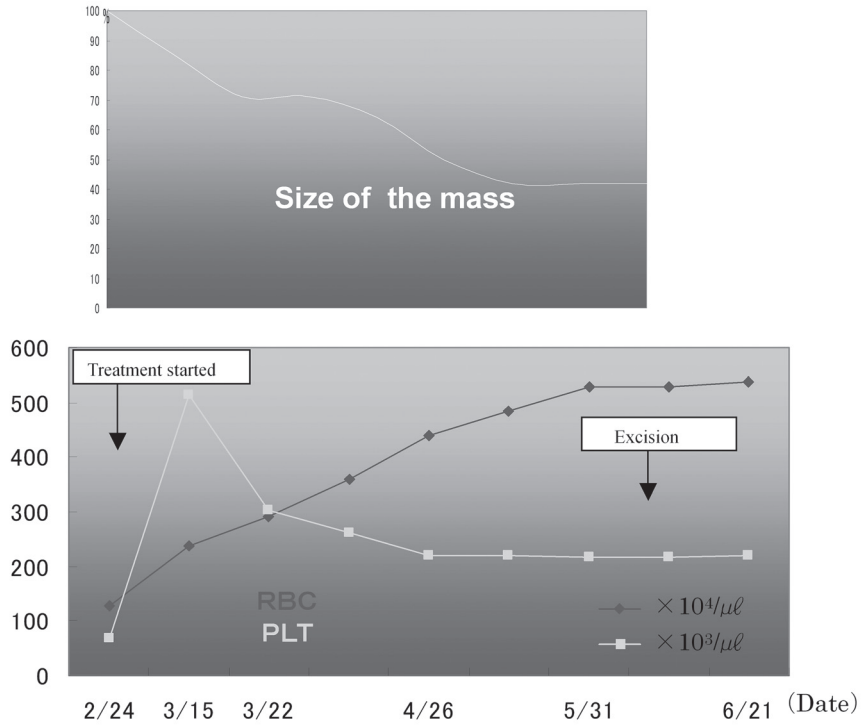


Figure2 Hematological findings and changes in the size of the mass



Figure3 Appearance of the lesion three months after internal treatment. The lesion is less raised and less erythematous than in Fig. 1.

The disease was identified as megaloblastic anemia. The patient was given priority internal treatment. Bone marrow examination showed megaloblastic changes. The patient was started on treatment for megaloblastic anemia, with 940 mg of sodium ferrous citrate per day orally and 100 μg of vitamin B₁₂ per day by intramuscular injection. After one month of treatment, hemoglobin improved to 10.4 g/dl. The clinical findings of the tongue markedly improved after ferric medicine and vitamin B₁₂ were administered. The mass reduced in size as progress was made toward recovery from anemia. Reduction of the mass was noted 7 days after vitamin B₁₂ was administered (Fig. 2). The mass had decreased by approximately one-half three months after treatment (Fig. 3). The remaining mass was resected.

Histopathological examination revealed a non-keratinized incassate epithelium, with irregular acanthosis without evident cellular atypia. Hyperplasia of the capillary vessels and collagen fibers was also identified without inflammatory cell infiltration to the adjacent lamina propria (Fig. 4). These findings were consistent with the diagnosis of so-called fibroma. The postoperative course was uneventful.

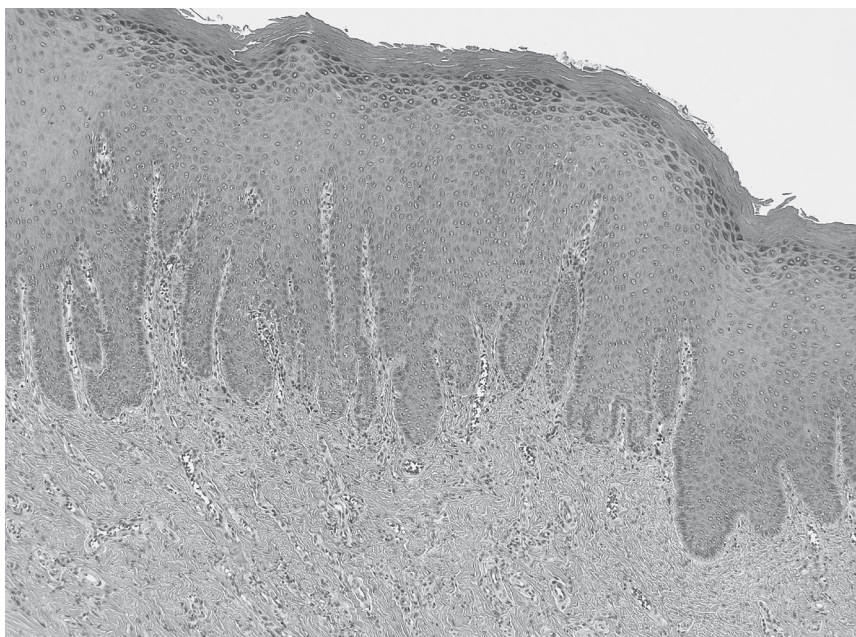


Figure4 Histological findings of the tongue mass.
Photomicrograph of the tongue mass showing no truly
paraneoplastic lesion and non-specific inflammatory hyperplasia
(Hematoxylin and eosin stain, $\times 100$)

Discussion

Glossitis and glossodynia are classic oral symptoms of pernicious anemia^{2,3,4,5,6}. The tongue is beefy-red and inflamed with small erythematous areas on the tip and margins. There is a loss of filiform papillae, and in advanced disease, papillary atrophy involves the whole tongue surface together with a loss of normal muscle tone. Following treatment with vitamin B₁₂, the tongue usually shows complete healing with resolution of the symptoms and reversal of the morphologic alterations⁷. Disturbances in taste can

occur and a decrease in salivation, eventually leading to xerostomia, may be present. Patients also may complain of dysphagia. Although the burning mouth sensation diagnosed in pernicious anemia can be due to neuropathy, a secondary fungal infection of the anemic mucosa by *Candida albicans* also has to be considered^{4,5)}. Changes in the oral mucosa, resembling those seen in pernicious anemia, may occur in such diseases as marked sideropenic anemia, folic acid deficiency, ariboflavinosis, malabsorption, and Sjögren's syndrome⁴⁾. In this case, his fissured tongue and tongue mass were not diagnostic of megaloblastic anemia.

Reports on oral lesions associated with megaloblastic anemia are extremely few. For tongue masses, no literature mentions an association with megaloblastic anemia. It is an interesting finding that his mass reduced with normalized serum levels after substitution therapy. General dentists need to be familiar with the clinical symptoms of this disorder.

REFERENCES

- 1) Chanarin I : The megaloblastic anemias, Oxford, Blackwell Scientific Publications, pp. 622-626, 1969.
- 2) Zegarelli DJ. Burning mouth : An alternative explanation for some patients with diabetes mellitus and pernicious anemia. *Ann Dent* 46 : 23, 1987.
- 3) Sugihara T, Yawata Y : Pernicious anemia. *Nihon-Rinsho* 50 : 771-786, 1992.
- 4) Millard HD, Gobetti JP : Nonspecific stomatitis – a presenting sign in pernicious anemia. *Oral Surg Oral Med Oral Pathol* 39 : 562, 1975.
- 5) Hjorting-Hansen E, Bertram U : Oral aspects of pernicious anemia. *Br Dent J* 125 : 266-270, 1968.
- 6) Field EA, Speechley JA, Rugman FR, Varga E, Tyldesley WR. Oral signs and symptoms in patients with undiagnosed vitamin B₁₂ deficiency. *J Oral Pathol Med.* 24 : 468-470, 1995.
- 7) Greenberg MS. Clinical and histologic changes of the oral mucosa in pernicious anemia. *Oral Surg Oral Med Oral Pathol* 52 : 38-42, 1981.

巨赤芽球性貧血に伴い舌に腫瘤を生じた1例

小佐野仁志^{1),2)} 宮内 徹²⁾ 外島 正樹³⁾
松本 浩一²⁾ 草間 幹夫²⁾

要 約

巨赤芽球性貧血は、VB12や葉酸の不足により骨髄造血細胞に核酸合成障害が起こり、細胞分裂が不完全となり、巨大な細胞が発生し、特異的な形態と機能障害により貧血を呈する疾患である。舌の腫瘤を主訴に当科を受診し検査の結果、巨赤芽球性貧血が合併したと診断された症例を経験した。巨赤芽球性貧血と舌腫瘤との関連性は低いと考えられるものの、舌の腫瘤が貧血の改善とともに縮小した興味ある症例を経験したので報告する。

初診の約4ヵ月前から舌背中央部に小豆大の腫瘤を自覚するも放置。しかし消失しなかった

ため近医内科を受診。経過をみていたが改善せず、増大傾向を認めたため当科を紹介された。血液一般検査より大球性貧血、汎血球減少を認めたため、血液内科に対診し、VB12欠乏性巨赤芽球性貧血の診断を得た。貧血の治療を優先し、VB12製剤の筋肉内注射を約7ヵ月間、鉄剤を約6ヵ月間投与したところ経時的に貧血状態の改善を認め、それとともに腫瘤の縮小を認めた。最終的に残存した腫瘤は切除した。術後の経過は、再発は認められず良好である。

(キーワード: VB12欠乏, 巨赤芽球性貧血, 舌腫瘤)

1) 自治医科大学附属さいたま医療センター 歯科口腔外科

2) 自治医科大学 歯科口腔外科学講座

3) 自治医科大学 内科学講座血液学部門