



## Case Report

# Management considerations for the patient with tongue squamous cell carcinoma associated with Behçet's disease: A case report

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## ABSTRACT

Malignancies associated with Behçet's disease (BD) are rare, and surgery for patients with BD frequently results in postoperative complications such as wound dehiscence, postoperative infection, and exacerbation of BD itself. We present a rare case of left tongue carcinoma in a 70-year-old woman with BD. The pathological findings showed moderate-differentiated squamous cell carcinoma with atypical squamous cell infiltrative proliferation. Daily concurrent chemoradiotherapy with docetaxel and cisplatin using superselective intra-arterial infusion via superficial temporal artery and occipital artery was performed. Two indwelling catheters, one from the left occipital artery to the left lingual artery and the other from the left superficial temporal artery to the left facial artery, were inserted to carry out superselective intra-arterial infusion treatment. After completion of all treatments, a biopsy specimen of the primary lesion showed grade III, nonviable tumor cells, suggesting complete response. Superselective intra-arterial chemoradiotherapy was effective for treating the tongue carcinoma associated with BD, and avoided surgical treatment. This method could control the primary lesion and cervical lymph node metastasis clinically without severe side effects. The patient has been disease free for 18 months without the complication of treatment.

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## 1. Introduction

Behçet's disease (BD), a syndrome that was first described by Behçet in 1937, is a multi-organ invasive refractory disease characterized by recurrent inflammatory manifestations, including oral aphthous ulcers, skin lesions, uveitis, and genital ulcers. Diagnostic criteria for BD were established by the International Study Group in 1990 [1]. Oral aphthous ulcer, the most frequent finding, is an important clinical diagnostic criterion of diagnosis.

The association of BD with malignancy is so uncommon that only 112 cases of malignancies in patients with BD, in the absence of oral cancer, have been reported to date [2]. Because BD is associated with a high incidence of postoperative complications such as wound dehiscence, postoperative infection, and exacerbation of BD itself [3], treatments that avoid surgery would be useful. No authors suggested any increase in chemotherapy toxicity, and no data about radiation therapy morbidity in the presence of BD in the literature [4].

In this report we report the use of superselective intra-arterial infusion chemoradiotherapy for the treatment of tongue carcinoma in a patient with BD. The primary tumor and neck lymph node metastasis showed complete response after treatment.

## 2. Case report

The patient was a 70-year-old woman with a painful and erosive lesion on the left margin of the tongue. She was diagnosed at 34 years as having BD accompanied by recurrent oral aphtha, genital ulcer, and erythema nodosum of the lower limbs. As these symptoms were mild, she underwent only observation without medication.

In the patient's more recent history, recurrent aphtha had been noted in the left margin of the tongue for the past 7 years. An aphthous ulcer with pain appeared 2 years ago, and pain was relieved by steroid ointment application. However, because the aphthous ulcer has recurred with increasing frequency over the last 6 months, the patient was referred to our department from dermatology.

At first visit, a 42 mm × 18 mm induration with ulcer was observed in the left margin of the tongue (Fig. 1), and a lymph node (about 10 mm) was palpable in the left submandibular region. The pathologic diagnosis of the lesion was moderately differentiated squamous cell carcinoma (Fig. 2). Enhanced

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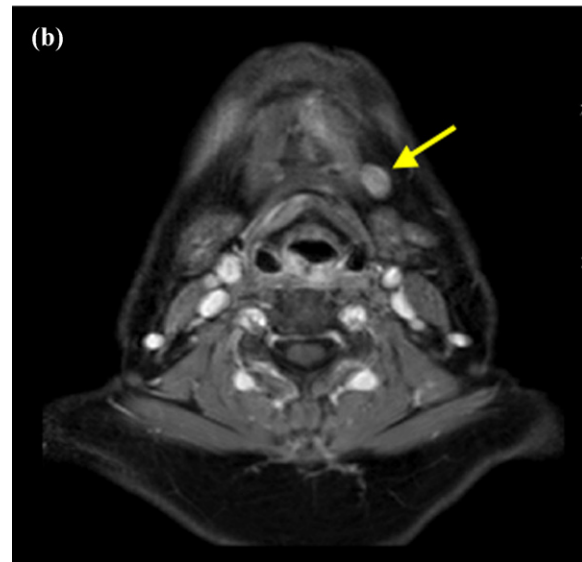
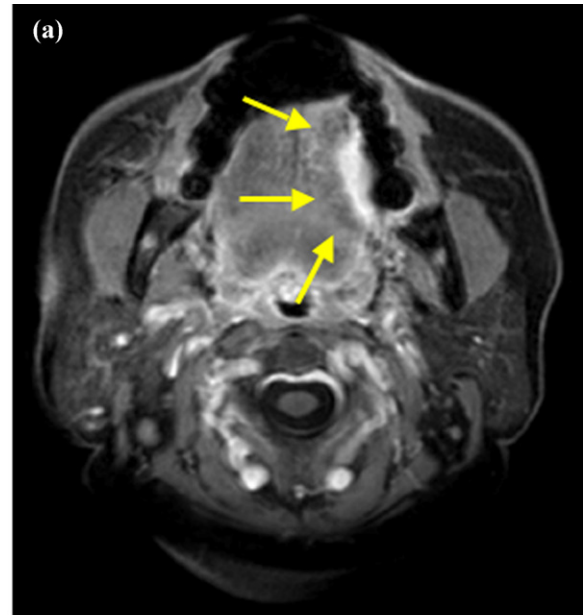


**Fig. 1.** A 42 mm × 18 mm ulcer with induration in the left margin of the tongue.

magnetic resonance imaging revealed a space-occupying mass (45 mm × 23 mm × 8 mm) in the left side of the tongue and pathologic lymph node enlargement (Fig. 3). The distant metastasis workup was negative. The patient was diagnosed as having tongue carcinoma T3N1M0 based on medical examination.

BD is associated with a high incidence of postoperative complications, and we performed superselective intra-arterial chemoradiotherapy. Two catheters were inserted superselectively into the left facial artery via the left superficial temporal artery and left lingual artery via the occipital artery [5,6]. The patient received daily concurrent chemoradiotherapy with docetaxel (DOC) and cisplatin (CDDP) via intra-arterial infusion. Each drug was given in equivalent doses from each catheter at the same time as radiotherapy was performed. DOC and CDDP doses were 10 mg/m<sup>2</sup>/week (total 50 mg/m<sup>2</sup>) and 5 mg/m<sup>2</sup>/day (total 120 mg/m<sup>2</sup>), respectively. External irradiation was performed 5 times per week at 2 Gy per fraction, up to a total of 48 Gy. The area of irradiation included the primary lesion and the neck lymph node metastasis. Grade 3 stomatitis and grade 3 neutropenia (National Cancer Institute common toxicity criteria, 2004) appeared during treatment.

The primary tumor and the lymph node metastasis were visualized with contrast medium on angiography computer tomography (Fig. 4). After completion of all treatments, the biopsy specimen from the primary lesion showed grade III, nonviable tumor cells (Fig. 5), and the primary tumor and regional lymph node metastasis in the neck were confirmed. At 18 months after treatment, the tongue surface became smooth, although with slight scar hard-

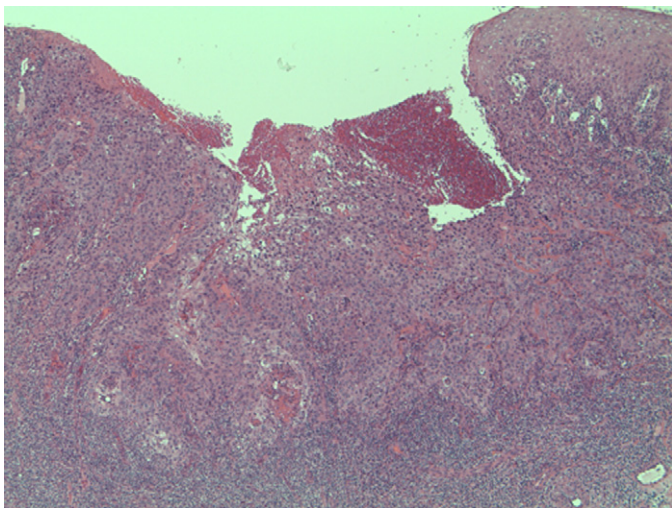


**Fig. 3.** (a and b) Magnetic resonance imaging shows pretreatment of the primary lesion in the left margin of the tongue and the lymph node in the left submandibular region (arrow).

ening (Fig. 6), enhanced magnetic resonance imaging there are no recurrence and metastasis (Fig. 7), and the treatment was deemed to have achieved complete response.

### 3. Discussion

In patients with BD, slightly more than 100 cases associated with malignancy have been documented. However, the direct relationship between BD and malignancy has not been proven [6]. The risk of cancer is increased in the presence of autoimmune diseases and vasculitides such as rheumatoid arthritis, progressive systemic sclerosis, Sjögren's syndrome, and systemic lupus erythematosus. For this reason, genomic and chromosomal instability, abnormalities in immune regulation such as T-cell deficiency, and overproliferation of B cells may trigger a malignant transformation of cells. Immunosuppressive drugs used for the treatment of autoimmune diseases are considered to induce malignant transformation in most cases [7].



**Fig. 2.** Histopathologic examination (hematoxylin and eosin stain, original magnification ×100) showing moderately differentiated squamous cell carcinoma with atypical squamous cell infiltrative proliferation.

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