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Case report

A case of subcutaneous panniculitis-like T-cell lymphoma of the cheek

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ABSTRACT

Subcutaneous panniculitis-like T-cell lymphoma (SPTL) was first reported as T-cell lymphoma resembling panniculitis. Many of the lesions are observed in the upper and lower extremities and trunk, whereas occurrence in the facial area is extremely rare. Our patient was a 19-year-old male who reported of some swelling in the left facial area 5 months prior to the initial examination. Originally misdiagnosed as a scar caused by accidental biting the buccal mucosa when first presenting at the Department of Dermatology of a local hospital, the patient was eventually referred to our department upon request for resection. Upon initial examination, a high degree of diffuse swelling extending from the left cheek to the eye socket was observed. Intraoral observation showed no pathological findings or tooth infections. MRI showed an unclear boundary in the subcutaneous fat layer of the left buccal region. Clinical and image findings with a cheek biopsy confirmed a diagnosis of Weber–Christian disease. The pathological diagnosis was SPTL. Chemotherapy resulted in disappearance of the swelling. The patient has remained relapse-free for 5 years.

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1. Introduction

Subcutaneous panniculitis-like T-cell lymphoma (SPTL), which was first reported as T-cell lymphoma resembling panniculitis, is a rare extranodal lymphoma mainly infiltrating the subcutaneous fatty tissue and has been classified as a type of cutaneous T-cell lymphoma [1,2]. Clinical features and image findings of SPTL are similar to those of panniculitis associated with autoimmunity; therefore, clinical diagnosis is particularly difficult. Although the incidence is very rare and there is no specific gender predilection, this disease is most often diagnosed in young people. Many of the lesions of SPTL are observed in the upper and lower extremities and the trunk, whereas occurrence in the facial area is extremely rare. Here we report our experience with a case of SPTL of the cheek in a teenage patient.

2. Case summary

The patient was a 19-year-old male who first visited our department with a chief complaint of swelling in the left cheek. The patient had a history of atopic dermatitis. The patient's current condition began 5 months before the first visit, when he reported becoming aware of some swelling in the left facial area. The patient was examined in the Departments of Otolaryngology, Plastic Surgery, Neurosurgery and Dermatology over the 5-month period, but the cause of the swelling remained unknown. No change in symptoms was observed for 5 months. Originally misdiagnosed as a scar caused by accidentally biting the buccal mucosa when first presenting to the Department of Dermatology, the patient was eventually referred to our department upon request for resection.

At that time, the patient's body temperature was 37.7 °C, and he reported mild fatigue. Extraoral findings showed asymmetry in facial appearance with a high degree of diffuse swelling extending from the left cheek to the eye socket. Although there was no redness, the skin in that area was elastic, tender; and warm to the touch. In addition, there was no movement disorder or paraesthesia of the facial muscles. The distance measured between the oral mucosa and skin of the cheek in the swelled region was 20 mm, whereas the distance of the same region on the opposite side was

* AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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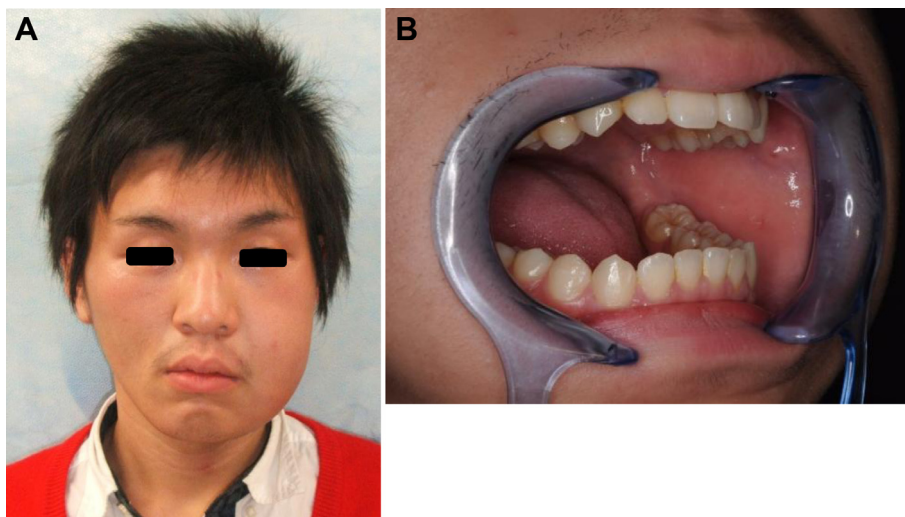


Fig. 1. Facial and intraoral findings during initial medical examination. (A) Facial photograph at the time of initial medical examination. Diffuse swelling was detected in the left cheek. (B) An intraoral photograph from the initial medical examination showing no abnormalities in the oral cavity.

7 mm (Fig. 1A). The submandibular and cervical lymph nodes were not enlarged.

Intraoral findings showed a mouth opening capacity of 60 mm with no disturbance in mouth opening. No swelling or redness was observed in the left cheek mucosa, and saliva outflow from the left parotid papilla was normal (Fig. 1B). Other intraoral observations showed no pathological findings or tooth infections.

Hematological findings showed a reduced WBC count of 2400 cells/ μ L and decreased liver function (AST level, 74 U/L; ALT level, 64 U/L; and LDH level, 387 U/L) (Table 1).

Panoramic X-ray showed an increased shadow throughout the left maxillary sinus. No other abnormal findings were observed.

As shown by MRI in Fig. 2A and B, the subcutaneous fat layer of the left buccal region had a low signal in the T1-weighted image and a markedly high diffuse signal in the T2-weighted image. Clinical findings suggested swelling of the subcutaneous tissue spreading from the cheek to the surrounding area. The findings of MRI showed significant thickening, and increased fat tissue were the main reasons for the swelling. Because no skin or dental infection

Table 1
Blood test results from the initial examination.

Peripheral blood (standard range)	Blood biochemistry (standard range)
WBC: 2400/ μ L (3500–9300/ μ L)	Alb: 4.2 g/dL (4.0–5.0 g/dL)
RBC: 541 $\times 10^4$ / μ L (400–557 $\times 10^4$ / μ L)	T-Bil: 0.6 mg/dL (0.3–1.2 mg/dL)
Hb: 13.9 g/dL (13.4–17.6 g/dL)	AST: 74 U/L (13–33 U/L)
Hct: 42.6% (38.9–50.7%)	ALT: 64 U/L (8–42 U/L)
Plt: 15.3 $\times 10^4$ / μ L (12–40 $\times 10^4$ / μ L)	LDH: 387 U/L (119–229 U/L)
	BUN: 15 mg/dL (8–22 mg/dL)
	CRE: 0.68 mg/dL (0.6–1.10 mg/dL)
	CRP: 0.22 mg/dL (0–0.39 mg/dL)

was observed, panniculitis was suspected. A clinical diagnosis of suspected Weber–Christian disease was made.

3. Symptoms and course

Although the etiology is unknown, Weber–Christian disease is reported as a cause of repeated and recurrent subcutaneous

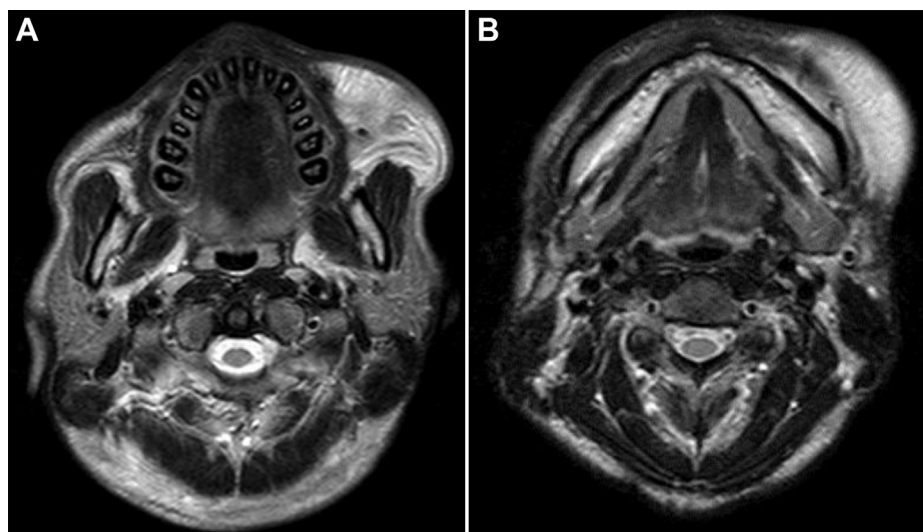


Fig. 2. T2-weighted MRI image (horizontal plane). (A, B) A markedly high signal is shown in the left buccal region.

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