CASE REPORT

Squamous cell carcinoma arising from lupus vulgaris with a >60-year history

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Received: 5 December 2016 / Accepted: 30 March 2017 / Published online: 9 April 2017
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Abstract We report the case of a 71-year-old Japanese man with squamous cell carcinoma arising from lupus vulgaris on the face, >60 years after the appearance of the lupus vulgaris. The red plaque on the patient's face had been diagnosed as a hemangioma or rosacea at several hospitals, although he had had lung tuberculosis at the age of 4 and his father died from lung tuberculosis at 38 years of age. Although lupus vulgaris was the most frequent clinical form of true skin tuberculosis until the 1960s, it has become rare since then. Malignant tumors are known to occur in individuals with lupus vulgaris, with a reported rate of 0.5–10.5%. In light of Japan's "graying society," tuberculosis is still an important disorder, and clinicians must remain aware of cutaneous tuberculosis.

Keywords Lupus vulgaris · Squamous cell carcinoma · *Mycobacterium tuberculosis*

Introduction

Lupus vulgaris is the most frequent clinical form of true skin tuberculosis. Clinically, lupus vulgaris shows dark red, irregular-surfaced erythema and plaque, and it is similar to discoid lupus erythematosus and cutaneous granulomatous disorders including sarcoidosis and longstanding hemangioma. The clinical diagnosis of lupus vulgaris

is difficult because it has become a rare disease in recent years, although it was common before the 1960s. Malignant tumors are known to occur in individuals with lupus vulgaris. In light of Japan's "graying society," tuberculosis is still an important disorder, physicians must remain aware of cutaneous tuberculosis. Here, we report a case of squamous cell carcinoma arising from lupus vulgaris with a decade-long history.

Case report

A 71-year-old Japanese man with an asymptomatic nodule surrounded by red plaque on his right cheek developed a 10-mm erythema at the age of 7 years, and the plaque gradually became enlarged and elevated. He had lived in Northeast China prior to the age of 4 years, and he had contracted lung tuberculosis there. His father died from lung tuberculosis at 38 years of age. The patient's red plaque had been diagnosed as a hemangioma or rosacea at several hospitals. One month before his visit to our hospital in 2013, a new nodule developed in the erythematous lesion on his right cheek. On clinical examination, a violaceous plaque with a scar measuring 16 × 12 cm was observed on the right cheek and lower jaw. There was a 3-cm red tumor with ulceration on the erythematous plaque (Fig. 1).

Histopathologically, in the new red nodule on his right cheek, the round- or spindle-shaped atypical cells proliferated to the lower dermis without keratinization (Fig. 2). The nodules of epithelioid granuloma were scattered in the surrounding dermis of this tumor, with lymphocytes and Langerhans giant cells, but no caseating necrosis. Kinyoun staining of the tissue was negative and a polymerase chain reaction assay of the tissue could not detect *Mycobacterium tuberculosis*, *M. avium*, or *M. intracellulare*. However, colonies developed on Ogawa medium after 8 weeks

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Fig. 1 The clinical findings included well-defined erythematoviolaceous plaque and an erythematous nodule with ulceration on the right cheek and right lower jaw of the patient, a 71-year-old Japanese man

Fig. 2 Histopathological findings of the squamous cell carcinoma (the area indicated by the circles) and granulomatous lesions of the surrounding area (the area indicated by the asterisks) (hematoxylin-eosin, original magnification $\times 12.5$, $\times 80$)

of cultivation, which was confirmed as *M. tuberculosis* by a DNA–DNA hybridization analysis. No causal organism was detected on cultures of sputum, bronchoscopic smear, urine, or stool.

Computed tomography revealed calcified lymph nodes in the mediastinum that were considered old lung tuberculosis. The hematological examination showed renal dysfunction, with a creatinine level of 1.63 mg/dL (normal range 0.6–1.2 mg/dL) and a high level of QuantiFERON® TB-Gold, 9.33 IU/mL (normal range <0.35 IU/mL).

Based on these findings, the patient's skin lesion was diagnosed as squamous cell carcinoma arising from very longstanding lupus vulgaris. The tumor was completely excised, and the patient's tuberculosis was treated with a regimen consisting of a combination of rifampicin, isoniazid, pyrazinamide, and ethambutol for 6 months. The skin lesions improved, leaving an atrophic red scar.

Discussion

Lupus vulgaris is one of the true skin tuberculosis types that commonly occur on the face. Although before the 1960s lupus vulgaris was the most frequent clinical form of true skin tuberculosis, it has since then become rare. Malignant tumors are known to occur in individuals with lupus vulgaris, with a reported rate of 0.5–10.5% [1]. Although squamous cell carcinoma is the most common form, to our knowledge, only four cases of squamous cell carcinoma arising from lupus vulgaris have been reported since 2000.

In the present case, it was 64 years until the development of the squamous cell carcinoma in the lupus vulgaris. In previous reports, the average period before the onset of such a malignant tumor in lupus vulgaris was 31 years, and the longest period before the development of squamous cell carcinoma from lupus vulgaris was 50 years [2]. Chronic inflammation is considered to be the mechanism underlying the pathogenesis of squamous cell carcinoma development from lupus vulgaris [3]. In light of Japan's "graying society," tuberculosis is still an important disorder, and thus dermatologists must remain aware of cutaneous tuberculosis—including lupus vulgaris—despite its rarity.

Compliance with ethical standards

Informed consent The patient provided a written informed consent for his case report to be published anonymously.

Conflict of interest The authors declare that they have no conflict of interest.

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