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BRIEF REPORT

Successful High-Dose Radiation Treatment for Chemo-Resistant Oral Squamous Cell Carcinoma in a Kindler's Syndrome Patient

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Dear Editor:

The patient was a 54-year-old female who had been diagnosed with Kindler syndrome[1]. She noticed vulnerable white moss and erosion on her lip, which was histologically diagnosed as squamous cell carcinoma (Fig. 1A, B). No metastasis was detected; however, the tumor inoperably extended to her lip, buccal mucosa, and hard palate. Those lesions were initially treated with a selective arterial infusion of peplomycin through bilateral retrograde catheter. Once the elevated lesions became flat, recurrence was observed within 2 months. Irrespective of sequential intra-arterial cisplatin chemotherapy commonly used for head and neck tumors treatment uplifting of the hard palate with fine granular white moss occurred (Fig. 1C). Computed tomography detected further invasion with bone destruction in the right maxillary sinus (Fig. 1D). As a third line treatment, we initiated intensity-modulated radiation therapy (IMRT) at a dose of 70 Gy/35 Fr (Fig. 1E). At one month after treatment, the tumor mass was significantly reduced in size and the oral lesions had noticeably improved. Followingly, the white elevated moss disappeared and magnetic resonance imaging could not detect the invasive lesion into the right maxillary sinus (Fig. 1F, G). No recurrence and distant metastasis have been observed 5 months after IMRT treatment.

Squamous cell carcinoma is reported to occur in nearly 70% of Kindler syndrome patients over 45 years of age, and the prognosis is reportedly poor due to the tumor agressiveness[2]. Recent report demonstrated that squamous cell carcinoma of the lip was successfully treated in 17 of 24 cases with selective administration of high-concentration peplomycin through the facial and maxillary artery[3]. However, the insufficient chemotherapy as the present case might be due to substantial tumor aggressiveness and/or phenotypic conversion to chemo-resistance.

Locally-intense radiotherapy was selected, because it high-dose irradiation could reportedly shrink oral squamous cell carcinoma arising in Kindler syndrome patients, irrespective of persistent mucosal disorder[4]. Based on recent development of irradiation devices, IMRT enabled selective and curative high-dose radiation therapy at the target sites with minimal adverse effects[5]. Differing from conventional irradiation methods, IMRT utilizes multiple radiation beams of non-uniform intensities which are modulated according to intensity maps resulting in strengthening the conformal radiation dose to the target lesions. In addition, particle beam therapy with stronger energy in comparison to radiation has been clinically applied in anticancer therapy. Although radiotherapy is known not to be commonly accepted for the treatment of genodermatosis patients due to toxicity and secondary carcinogenesis, high dose IMRT without redundant irradiation to

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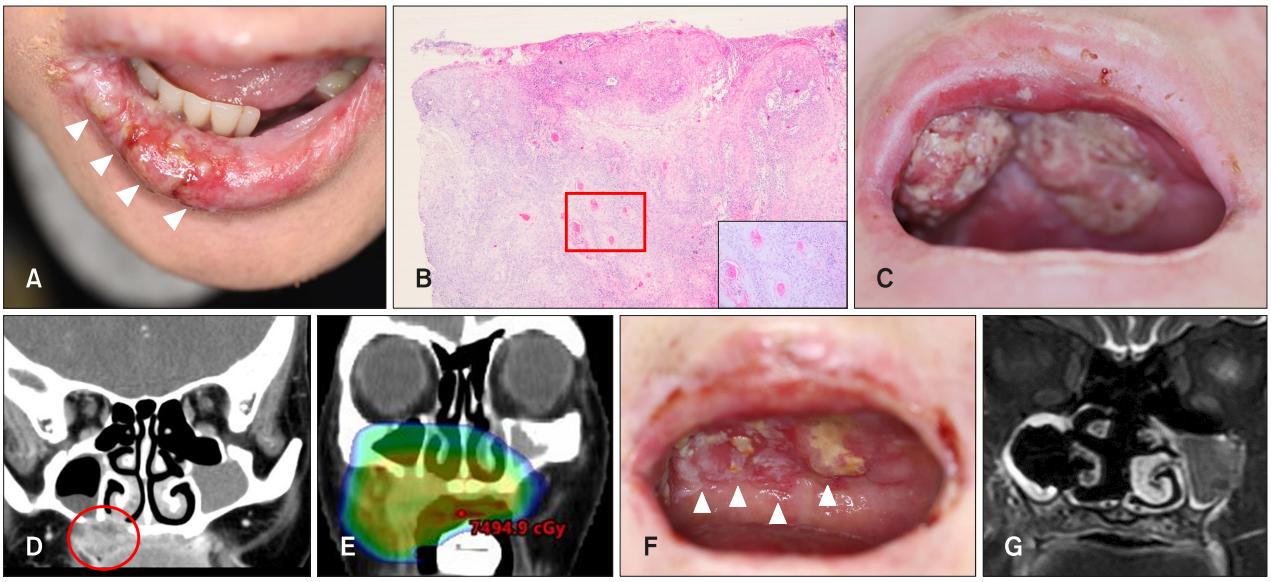

Fig. 1. (A) Vulnerable white moss and overlying erosion on the right side of the lip (arrowheads). (B) H&E staining demonstrated atypical squamous cells with the formation of cancer pearls deeply proliferated from the epithelium on the upper portion (×20). The inset shows a high-powered view of the area in the red rectangle (×200). (C) The elevated lesion with white moss on the hard palate was apparent. (D) Direct tumor invasion into the right maxillary sinus and nasal cavity through the bottom of the maxillary bone in a view of the coronal section (red circle). (E) A map of intensity-modulated radiation therapy (IMRT) irradiation indicating that the right eye ball was out of focus. The rainbow map was designed as non-uniform irradiation area. Total estimated dose on the center was 7,494.9 cGy. (F) The white elevated moss disappeared and was replaced by slightly necrotic tissue (arrowheads). (G) The tumor was not detected by magnetic resonance imaging after IMRT treatment.

the surrounding tissue was effective and well-tolerable in the present case. Since the follow-up period is relatively short as 5 months, further care should be taken to avoid the possible occurrence of secondary malignancies in the future. This experience will add significant new evidence on the treatment of chemo-resistant squamous cell carcinoma of the oral cavity arising in genodermatosis patients. We received the patient's consent form about publishing all photographic materials.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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Drug-Induced Immune Hemolytic Anemia Caused by Postoperative Cefotetan Administration

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Dear Editor:

Drugs can induce almost any hematologic disorder that affects white blood cells, red blood cells (RBCs), platelets, and the coagulation system. Although the clinical manifestation of drug-induced immune hemolytic anemia (DIIHA) is usually mild, it may progress to acute severe hemolytic anemia (HA) and death. There is an increasing number of reports of second and third-generation cephalosporins causing clinical hemolysis[1]. Cefotetan is a second-generation cephalosporin used frequently in postoperative condition. Here, we report a case of cefotetan-induced immune hemolytic anemia in a Korean (CIIHA).

A 68-year-old male with a small nasal nodule was diagnosed as basal cell carcinoma was referred to our hospital for further treatment. We removed the remaining lesion and the defect was repaired with a nasolabial flap (Fig. 1). Intravenous cefotetan 2 g daily was administered to the patient postoperatively for 7 days. Before discharge, the patient developed sudden mild fever (37.8°C) and fell down due to dizziness. Other symptoms including nausea, weakness, dark urine color, drowsiness, and pale appeared successively. During his hospitalization, there were no symptoms other than stomach discomfort. His hemoglobin (Hb) and hematocrit were 3.5 g/dl and 12.8%, respectively. The total serum bilirubin was 6.86 mg/dl (Table 1). There was no evidence of internal bleeding upon physical examination. His reticulocyte and red cell production index were 2.93% (normal, 1%～2%) and 0.916. We excluded chronic anemia, and acute gastrointestinal bleeding as a possible diagnosis. Therefore, we doubted a drug-induced condition. The patient was transferred to the Medical Intensive Care Unit. The patient's serum did not react with the antibody-detection RBCs. The

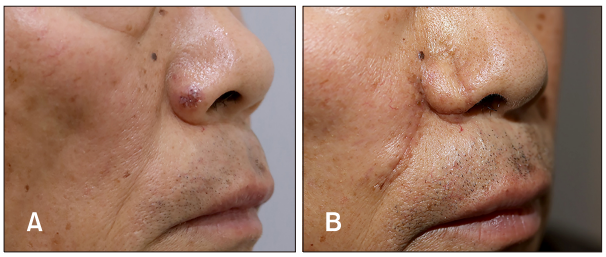

Fig. 1. A 1.0 cm×1.0 cm sized erythematous nodule with brown pigments on the nose (A) and at the 3 months follow-up (B). We received the patient's permission about publishing all photographic materials.

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