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Merkel-type oral small cell neuroendocrine carcinoma as second malignancy of tongue

KEYWORDS
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Small cell neuroendocrine carcinoma (SNEC) is an aggressive malignancy with less than 20 cases reported in the oral cavity.[1–5] Most cases were diagnosed at 6th to 8th decades with a male predilection. In the head and neck area, SNEC occurs most frequently in the larynx followed by the sinonasal cavity and salivary glands.[1] The clinical features of oral SNEC are nonspecific, which may appear as an ulcerated mass similar to other mucosal carcinomas. The tumor usually progresses rapidly and metastasis to cervical lymph nodes is common.

A 56-year-old male patient had history of squamous cell carcinoma of right ventral tongue and floor of mouth. The neoadjuvant chemotherapy, wide excision and post-surgical radiation therapy were performed seven years ago. After treatment, he was under a regular follow-up every three months. In the last follow-up, the patient complained dysphagia and slurred speech for one month. Two exophytic masses were present over the posterior dorsal tongue (Fig. 1A) and right lateral border of tongue (Fig. 1B) respectively. The masses revealed ulcerated surface, irregular borders and firm in consistency. The clinical examination revealed no cervical lymphadenopathy, and the tentative diagnosis was a second primary squamous cell carcinoma. Then the incisional biopsy of the dorsal tongue lesion was performed. Microscopically, it showed sections of nodular lesion covered by ulcerative surface (Fig. 1C) with aggregation of small, basophilic cells in the stroma. The tumor cells showed high nuclear to cytoplasmic ratio, and hyperchromatic, pleomorphic, round to oval nuclei (Fig. 1D). Abundant mitotic figures and confluent foci of necrosis (Fig. 1D, arrowheads) were also identified. Immunohistochemically, the tumor cells showed punctate paranuclear staining with cytokeratin (CK) (AE1/AE3) (Fig. 1E) and CK20 (Fig. 1F). The tumor cells were also positive for the neuroendocrine markers, including CD56 (Fig. 1G), synaptophysin (Fig. 1H) and chromogranin (Fig. 1I). The histopathological diagnosis was a Merkel-type oral small cell neuroendocrine carcinoma. Subsequently, the patient underwent several general surveys including oral and chest computed tomography, abdominal magnetic resonance imaging, esophagogastroduodenoscopy and general bone scan. The results revealed suspicious metastatic lymphadenopathy in left upper mediastinum and a metastatic lesion in the liver. He was under chemotherapy with cisplatin and etoposide, and the size of oral lesion decreased continuously during the regular follow-up; however, he expired from septic shock and respiratory failure six months after diagnosis.

Unlike the favorable prognosis of Merkel cell carcinoma (MCC) in skin, the MCCs of oral cavity revealed poorer prognosis.[1] Different to distinct MCC of skin or small cell carcinoma of lung, the SNECs of oral cavity consist of Merkel-type [CK20+/thyroid transcription factor-1(TTF-1)-] and pulmonary-type (CK20-/TTF-1+) according to their immunohistochemical expression pattern.[2] In our case, the SNEC of tongue presented as second primary malignancy of oral cavity seven years after surgery and radiotherapy for squamous cell carcinoma. These clinical features are

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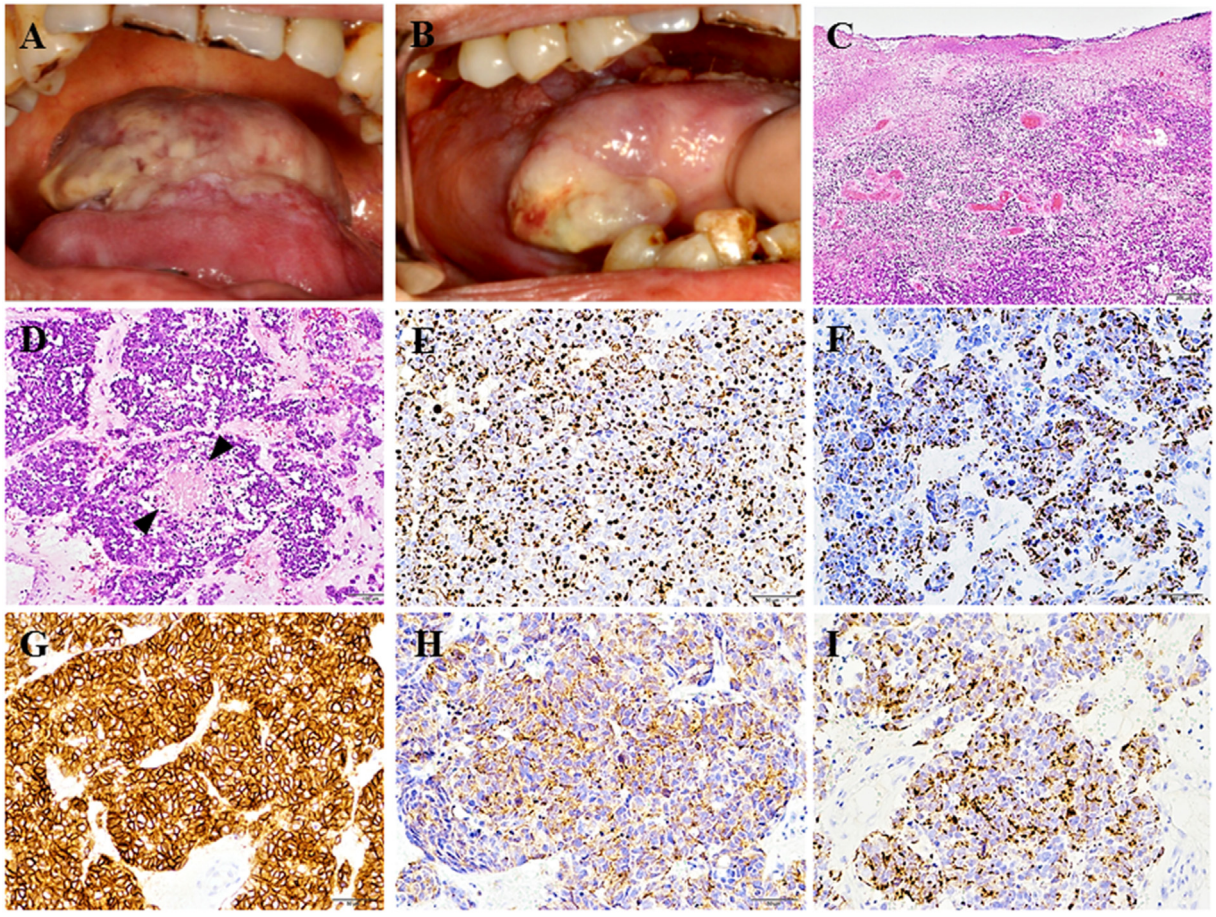

Figure 1 Clinical, and microscopic photographs of Merkel-type oral small cell neuroendocrine carcinoma as second malignancy of tongue. Ulcerated exophytic masses over the posterior dorsal tongue (A) and right lateral border of tongue (B). (C) The lesion shows a nodular lesion covered by an ulcerative surface (hematoxylin and eosin stain; magnification, 40×). (D) The tumor cells showed abundant mitotic figures, high nuclear to cytoplasmic ratio, and hyperchromatic, pleomorphic, round to oval nuclei, and confluent foci of necrosis (arrowheads) (hematoxylin and eosin stain; magnification, 100×). The tumor cells showed punctate paranuclear staining with cytokeratin (CK) (AE1/AE3) (E, magnification, 200×), CK20 (F, magnification, 200×) and also positive staining for CD56 (G, magnification, 200×), synaptophysin (H, magnification, 200×), and chromogranin (I, magnification, 200×).

resemble to the case previously reported by Jha at al.[5] SNEC of tongue is extremely rare, which implies that the SNEC of tongue may be a radiation-induced cancer. More studies are necessary to verify this hypothesis.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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