REPORT

Small cell neuroendocrine carcinoma (SCNEC) of the tongue: A case report

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Abstract: Small cell neuroendocrine carcinoma (SCNEC) of the tongue is very rare. We here present a SCNEC in a patient with distant metastasis. A 74-year-old Chinese male went to hospital to treat a tongue tumor, which was founded at a conventional physical examination in Weifang Stomatology Hospital. The check of positron emission tomography-computer tomography (PET-CT) by Weifang People’s hospital revealed a tumor in the right root of tongue, and distant metastasis in the right submandibular area, neck, mediastinum, right hilar, abdominal, retroperitoneal multiple lymph nodes, left thyroid, right lower lung, right scapula and bilateral adrenal. The patient was diagnosed tongue SCNEC by the pathological analysis of the tissue section. Conforming to the diagnosis of tongue SCNEC, the patient received adjuvant chemotherapy for 6 cycles with etoposide and carboplatin, and is alive now 9 months after the diagnosis.

Keywords: SCNEC, distant metastasis, chemotherapy, etoposide, carboplatin.

INTRODUCTION

SCNEC was first described by Travis in the lung (Travis et al., 1991), but cases of SCNEC were reported in other organs like uterus, thymus, stomach, kidney, bile duct, parotid gland, prostate, larynx and cervix (Iso et al., 2009; Kaminski et al., 2003; Yun et al., 1999; Evans et al., 2006; Mills., 2002). Most of the few reports of SCNEC in the head and neck were focused on the parotid glands, submandibular glands, etc. Most malignant tumors in the tongue are squamous cell carcinoma, we found cases of tongue SCNEC in the literature (Baker et al., 1999; Kusafuka et al., 2009; Yoshida et al., 1995). In this case report, we describe a rare case of tongue SCNEC with distant metastasis, the patient did not receive surgery and radiotherapy, received only six cycles of chemotherapy with etoposide and carboplatin. Because of the scarcity of such cases, the purpose of we report this case, expecting to provide reference for the treatment of such diseases and hoping to find the probably best clinical treatment on the SCNEC.

Case presentation

A 74-year-old Chinese male went to hospital to treat a tongue tumor, which was founded at a conventional physical examination. Computer tomography scan confirmed the presence of the tumor and revealed an about 4 cm tumor in the right root of the tongue, the tumor is presented in fig. 1. The check of positron emission tomography-computer tomography (PET-CT) by Weifang People’s hospital revealed the distant metastasis in the right submandibular area, neck, mediastinum, right hilar, abdominal, retroperitoneal multiple lymph nodes, left thyroid, right lower lung, right scapula and bilateral adrenal, the results of the PET-CT are presented in the fig. 2. Considering the patient had end-stage cancer, after communicated with the patient and family members, we cut out part of the tumor in the root of tongue for pathological examination.

Fig. 1: Computer tomography scan confirmed the presence of the tumor and revealed an about 4 cm tumor in the right root of the tongue.

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Microscopically, cancer cells distributed diffusely, showing solid sheet, partly of a cord-like, nuclear stained deeply, some cells had angular, nuclei was unclear, cytoplasm was rare and basophilic, and some nucleus were naked, which are presented in the fig. 3.

**Fig. 2**: PET-CT revealed the distant metastasis in the right submandibular area, neck, mediastinum, right hilar, abdominal, retroperitoneal multiple lymph nodes, left thyroid, right lower lung, right scapula and bilateral adrenal.

**Fig. 3**: Cancer cells distributed diffusely, showing solid sheet, partly of a cord-like, nuclear stained deeply, some with angular, fine chromatin was dusty, nuclei was unclear, cytoplasm was rare and basophilic, and some nucleus were naked (Hematoxylin and eosin staining H&E 20x).

Immunohistochemical analysis demonstrated that the CD56 and synaptophysin were obviously positive in the cancer cells and presented in the fig. 4 and fig. 5, focally positive of the chromogranin and TTF-1, which is presented in the fig. 6. The proliferation index, evaluated with Ki-67 was 80%. According to the radiologic and pathologic findings (Shin et al., 2000), the patient was diagnosed with neuroendocrine carcinoma of small cell (SCNEC) on the tongue. The patient was then treated with etoposide and carboplatin for 6 cycles without surgery.

**RESULTS**

The patient is alive now, nine months after the diagnosis, the current life cycle has been more than 9 months, and there is no significant deterioration in general condition, we will keep focusing on the situation of the patient.

**DISCUSSION**

Tongue small cell neuroendocrine carcinomas are very rare. According to the new classification by the WHO (Capella et al., 2000), the neuroendocrine tumors are divided into two kinds, including the well differentiated...
tumors (carcinoids) and poorly differentiated neuroendocrine carcinoma (small cell carcinoma). Both the number of the two kinds patients are very poor, together with the lack of the standardized treatment shame, the poor clinical experience, and the limited literature, all above-mentioned factors make it to be a huge challenge for the doctors. More researches are warranted to find the best clinical treatment of tongue SCNEC. This patient tumor was firstly found in the tongue, but more organs including the lung were invaded by the tumor from the check of PET-CT, and the patient refused the further examination, made it difficult to conform the primary organ, but either primary or secondary SCNEC is very rare.

Fig. 6: Tumor cells showed diffuse positivity for TTF-1. The nucleus was colored.

This patient's PET-CT examination reported the multiple distant metastases in the right submandibular area, neck, mediastinum, right hilar, intraperitoneal, retroperitoneal, and suggested that lymphatic metastasis is an important pathway for SCNEC.

According to the previous reports, the therapy for tongue SCNEC is surgery alone or surgery combined with chemotherapy (Mineta et al., 2001). Considering the emergence of multiple metastases of the patient, there was no surgery indication (Takei, et al., 2002). Based on pathological diagnosis, the patient received 6 cycles of chemotherapy with etoposide and carboplatin.

CONCLUSION

The etoposide and carboplatin still have therapeutic significance to the later period patient of SCNEC, even the patient has the distant metastasis. The both drugs can extend the life of the SCNEC patient, and improve the quality of patient’s life.

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REFERENCES

Travis WD, Linnoila I, Tsokos MG, Hitchcock CL, Cutler GB and Nieman L (1991). Neuroendocrine tumors of
