Metastatic neuroendocrine carcinoma to the mandibular gingiva from the duodenum papilla

ABSTRACT

Neuroendocrine carcinoma is a high-grade carcinoma with morphological and immunohistochemical features of neuroendocrine differentiation. Poorly differentiated neuroendocrine carcinoma which has two subtypes (small cell or large cell type) is extremely rare, especially in head and neck cancers. Metastatic neuroendocrine carcinoma to the oral and maxillofacial region is even rarer. We report a rare case with metastatic neuroendocrine carcinoma to the mandibular gingiva from the duodenum papilla. Surgeons may consider metastatic tumors as well as primary tumors, because neuroendocrine tumors are extremely rare in oral and maxillofacial region.

1. Introduction

Neuroendocrine tumors arise commonly in the lung and gastrointestinal tract. Neuroendocrine carcinoma is a high-grade carcinoma with morphological and immunohistochemical features of neuroendocrine differentiation. Poorly differentiated neuroendocrine carcinoma which has two subtypes (small cell or large cell type) is extremely rare, representing 0.3% of head and neck cancers [1]. Furthermore, metastatic neuroendocrine carcinoma to the oral and maxillofacial region is even rarer. We report a rare case of metastatic neuroendocrine carcinoma to the mandibular gingiva from the duodenum papilla.

2. Case report

A 73-year-old female was referred to our department with a left painful mandibular gingival mass and numbness to the left chin and lip. The 30 × 25 × 20 mm mass was elastic hard and had indentation caused by maxillary teeth (Fig. 1A). Computed tomography showed multiple cervical, mediastinal and paraaortic lymph node metastases as well as a left mandibular tumor (Fig. 1B–C). Four months before the first visit to our department, the patient had undergone pancreaticoduodenectomy for poorly differentiated small cell neuroendocrine carcinoma of the duodenum papilla. The pathological examination of the duodenal papilla revealed diffuse proliferation of chromatin abundant atypical cells in hematoxylin and eosin staining (Fig. 1F). The tumor cells expressed immunoreactivity for CD56, synaptophysin, and AE1/AE3. The mandibular gingival biopsy was performed, and diffuse proliferation of chromatin abundant atypical cells was shown in hematoxylin and eosin staining (Fig. 1F). Therefore, the mandibular gingival tumor was diagnosed as metastatic neuroendocrine carcinoma. The patient with further multiple liver metastases and peritoneal dissemination underwent palliative treatment but died 1 month after the first visit to our department.

3. Discussion

Metastasis to the oral and maxillofacial region is reported to account for 1–1.5% of all oral and maxillofacial malignancies [2,3]. The most common location was the mandible (44.9–51.3%), followed by gingival soft tissue (16.8–23.3%) [2,3]. Paresthesia or anesthesia to the lip and chin can be described as numb chin syndrome, which is usually unilateral and affects the inferior alveolar nerve in the mandible [4]. The syndrome commonly raises the suspicion of primary or metastatic disease to the mandible. In the present case, the metastatic oral site was mandibular gingiva, and numb chin syndrome was shown.

In systematic reviews of metastasis to the oral and maxillofacial region, the most usual primary site was the lung in men (14.1–20.5%) and the breast in women (11.5–31.1%) [2,3]. In 70% of patients with metastatic oral malignancy, primary site was diagnosed before the metastatic site, while the metastatic site was discovered first in 30% of patients [2]. Over 40% of pancreatic neuroendocrine tumors become metastatic in the course of the disease, commonly by lymph nodes or liver metastasis [5]. Metastasis to the oral and maxillofacial region from the pancreas was seen in an incidence of 0.6–0.9% in recent reviews, whereas metastatic
oral tumors did not show sufficient histopathological information [2,3].

To our knowledge, present case was the first report of oral metastasis from the duodenum papilla. Surgeons may consider metastatic tumors as well as primary tumors, because neuroendocrine tumors are extremely rare in oral and maxillofacial region.

Declaration of competing interest

None declared.

References


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Fig. 1. Intraoral photograph, computed tomography (CT), and Histopathological photographs. A: Intraoral view showed an elastic hard mass of the left mandibular gingiva, B, C: CT showed a left mandibular tumor (Arrow) and multiple cervical lymph node metastases. D,E: Pathological examination of the duodenal papilla revealed diffuse proliferation of chromatin abundant atypical cell in hematoxylin and eosin staining and MIB-1 index >95%, F: Pathological examination of mandibular gingival tumor showed diffuse proliferation of chromatin abundant atypical cell in hematoxylin and eosin staining.