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## Correspondence

# Submandibular gland metastasis as the initial manifestation of lung small cell carcinoma

## KEYWORDS

Lung;  
Metastasis;  
Neuroendocrine carcinoma;  
Small cell carcinoma;  
Submandibular gland

Neuroendocrine carcinomas arise commonly in the lung and gastrointestinal tract. 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.<sup>1</sup> Distant metastases of lung small cell carcinoma mainly occur in the liver, adrenals, bones, or brain,<sup>2</sup> and the metastasis to the submandibular gland (SMG) is extremely rare.<sup>2,3</sup> Here, we reported a rare case of SMG metastasis as the initial manifestation of lung small cell carcinoma.

A 61-year-old male visited our hospital for a left submandibular mass and facial nerve palsy (Fig. 1A). The mass without mobility was elastic hard. Computed tomography showed the 35 × 34 × 30 mm left heterogeneous SMG (Fig. 1B). A 38 × 26 mm lobular mass was found in the upper lobe of the left lung, and a 50 × 35 mm mass in the left pulmonary hilum with indistinct boundaries with the pulmonary artery was suggestive of lymph node metastasis (Fig. 1C and D). Magnetic resonance imaging showed a 35 × 32 × 30 mm heterogeneous tumor in the left SMG, and multiple nodules with enhancing effects in the cerebellum, midbrain, cerebrum, and pineal gland. [(18)F]-fluorodeoxyglucose (FDG) positron emission tomography revealed FDG accumulations of left SMG, left upper lung lobe, hilar region, and right adrenal gland (Fig. 1E). The SMG biopsy showed small tumor cells arranged in sheets and were separated fibrous stroma, and tumor cells stained

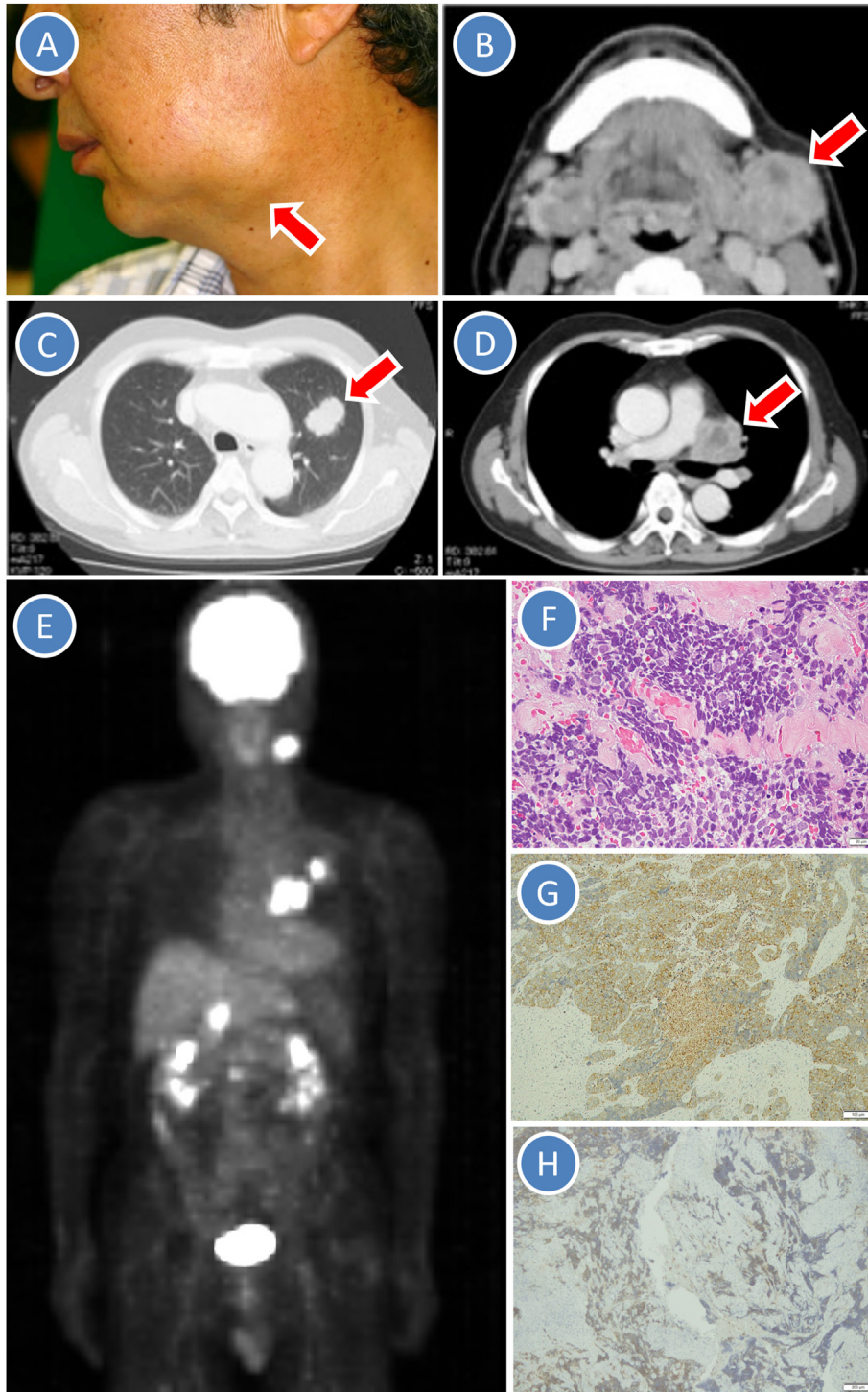
positively for synaptophysin, CD56, chromogranin A, CK AE1/AE, and CK CAM5.2 (Fig. 1F–H). The SMG lesion was diagnosed as neuroendocrine carcinoma. Because primary lung cancer was suspected by imaging findings and pathological diagnosis of the SMG, a transbronchial lung biopsy was performed in the Department of Respiratory Medicine and the lung lesion showed similar pathological findings to the SMG lesion. The final diagnosis was lung small cell carcinoma with multiple systemic metastases. The patient received 2 courses of chemotherapy (cisplatin and irinotecan) and palliative irradiation of the brain and submandibular region. Supportive care was provided for progressive disease, but the patient died 7 months after the first visit to our department.

Metastasis in the oral and maxillofacial region is reported to account for 1–1.5% of all oral and maxillofacial malignancies,<sup>4,5</sup> and the most usual primary site was the lung (about 20%).<sup>4,5</sup> Recent reviews showed metastasis to the submandibular region from the lung was seen in an incidence of 0.6–0.9%, whereas metastatic oral tumors did not show sufficient histopathological information.<sup>4,5</sup>

Primary or metastatic neuroendocrine carcinomas of the SMG are extremely rare,<sup>2,3</sup> and the symptom is characteristically seen as an asymptomatic, palpable mass.<sup>2</sup> However, the tumor spread from the SMG may cause facial nerve palsy.<sup>2</sup> In the present case with submandibular mass and facial nerve palsy, SMG metastasis was the initial

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**Fig. 1** Extraoral photograph, radiological images, and histopathological photomicrographs. (A) The extraoral view showed a left submandibular mass (arrow). (B) Computed tomography (CT) showed a left submandibular gland tumor (arrow). (C) CT revealed a lobular mass (arrow) in the upper lobe of the left lung. (D) CT showed a lymph node metastasis (arrow) in the left pulmonary hilum. (E) [(18)F]-fluorodeoxyglucose (FDG) positron emission tomography revealed FDG accumulations in the left SMG, left upper lung lobe, hilar region, and right adrenal gland. (F) Pathological examination of the submandibular gland revealed small tumor cells arranged in sheets and were separated fibrous stroma (Hematoxylin and eosin stain). (G) Tumor cells stained positively for synaptophysin (Immunostaining). (H) Tumor cells stained positively for CD56 (Immunostaining).

manifestation of lung small cell carcinoma. When surgeons encountered neuroendocrine carcinomas of the SMG, the

metastasis as well as primary cancer of the SMG should be considered for the rarity.

### Declaration of competing interest

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

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