



Late pulmonary metastasis of submandibular adenoid cystic carcinoma

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Adenoid cystic carcinoma (ACC) is a rare epithelial tumor with a distinct natural history characterized by an indolent but persistent growth, late onset of distant metastases and eventual death of patients. We present a case with submandibular ACC metastatic to the lungs 14 years after primary resection. A 41-year-old woman was admitted to our hospital because of nodular lesions in both lungs that was detected incidentally by a chest radiograph. Chest CT scan revealed multiple nodular opacities of sizes ranging from 3 mm to 20 mm. Work up including CT scan of the abdomen, bone scintigram, brain MRI, endoscopy, gynecologic examination, examination of thyroid and breast, and other blood tests, the findings of which were normal. Fourteen years previously, the patient underwent a hemimandibulectomy for ACC of the submandibular gland. Thereafter, careful follow-up spanning 5 years revealed no lesions believed to be recurrence. The histological specimen from the nodular opacities, which was obtained by CT-guided percutaneous biopsy, was similar to that of the original ACC of the submandibular gland. Because the patient did not want aggressive therapy, no systemic chemotherapy was offered. She died of progression of the pulmonary lesions 4 years after the recurrence.

A generalization accepted in the treatment of cancer assumes that a cure is present if there is freedom from disease for 5 years. However, ACC of the submandibular gland is a slow-growing, insidious lesion with a prolonged natural history that includes a tendency toward local recurrence and distant metastasis (1-4). Sakes et al reported that disease recurrence, either loco-regional or metastatic occurred up to 156 months after radiotherapy (2). van der Wal (3) reported that the average time between the diagnosis of the primary lesion and the detection of metastases was 36.8 months (median 28.5 months); in the case of lung metastases the average time was 38.4 months (median, 45 months). Recently, Cohen et al reported that the time interval between initial therapy and distant metastasis ranged from 6 months to 77 months (median, 27 months) (4). Therefore, the concept of cure in patients with ACC of the submandibular gland may be difficult to assess until the patient is disease free for 10 years or longer (4). However, recurrence of ACC of the submandibular gland 14 years after the initial therapy in our patient is unique, and this represents one of the longest periods recorded as a case report. ACC of the submandibular gland frequently recurs with lung metastases, often asymptomatic (2-4). In addition, it is also interesting to note that pulmonary metastases are almost never solitary. When one or more are appreciated initial, other nodules sometimes appear very slowly as indicated by survivals in excess of 10 years (5). It should be recognized that follow-up needs to be long term to detect all late recurrence and that complete resection does not always mean cure. ACC of the submandibular gland represents a life-long threat to some patients and requires constant vigilance by medical practitioners.

Conflict of interest

We declare no conflicts of interest.

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