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Delayed pulmonary metastasis of adenoid cystic carcinoma originated from external earway: Case report

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ABSTRACT

Pulmonary metastases in a 68-year old male was detected 20 years after the operation for adenoid cystic carcinoma originated from external earway. Adenoid cystic carcinoma metastases was confirmed by fine-needle aspiration biopsy from multiple mass lesions in right chest. The pulmonary metastasis of adenoid cystic carcinoma emerging from external earway after such a long period is a rare case in the literature.[Turk J Cancer 2005;35(2):93-95].

KEY WORDS:

External earway, adenoid cystic carcinoma, pulmonary metastases, delayed metastases

INTRODUCTION

Adenoid cystic carcinoma of external earway originating from sweat glands is a rare tumor. The tumor generally has a benign clinical trend and emerges as crust verrucose discs or deep seated nodules. Detecting of dermal nodules and consisting of cystic area with solid fields is the main histological pattern. It has been reported that the presence of excessive solid fields is correlated with poor prognosis (1). Multiple organ metastasis may be detected in aggressive tumors (2). Although there is no consensus on treatment, surgery and postoperative radiotherapy might be preferred according to the degree of tumor invasion. It has been reported that the five year survival rate with the treatment is 61%, however this rate can be modified by the factors like histological grade, degree of invasion, and tumor existence in surgical margins (3). A rare case with delayed pulmonary metastasis who had been operated 20 years ago due to adenoid cystic carcinoma of external earway was reported here.

CASE REPORT

Multiple metastatic nodules were seen in right hemithorax on the X-ray taken for determining the etiology of thoracal pain in a 68-year old male. The patient underwent radical external earway excision + partial parotidectomy + radical neck dissection (there was not malignant involvement in parotid gland or lymph nodes) for adenoid cystic

carcinoma of external earway 20 years ago (Figure 1). He had also a history of anterior resection 10 years ago due to rectal cancer. In thoracal CT, multiple metastatic nodules were seen in right hemithorax at different size and localization (Figure 2). There was no mass lesion on bronchoscopy. Adenoid cystic carcinoma was confirmed by CT guided fine-needle aspiration biopsy (Figure 3). There was not another organ involvement in screening. Chemotherapy was started to the patient for metastatic adenoid cystic carcinoma. Disease remained stabilized after six cycles of gemcitabine (1000 mg/m^2 on days 1,8,15) and 5-fluorouracil (325 mg/m^2 for 5 days). The patient died because of acute myocardial infarction in his one year follow-up.

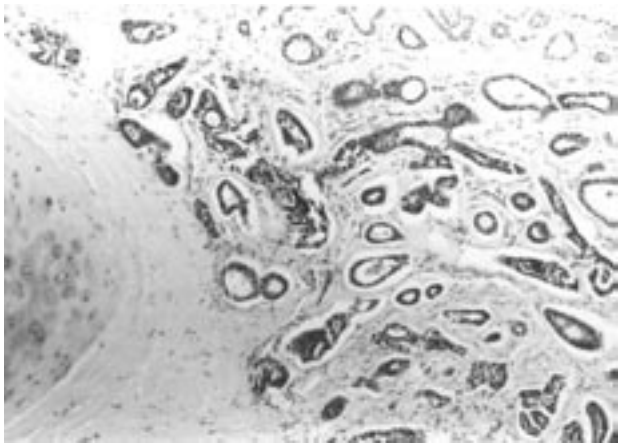

Fig 1. Adenoid cystic carcinoma invading cartilage. Tumor composed of nests and columns of cells of uniform appearance, arranged concentrically around gland like spaces containing hyaline material (H&E, x50)

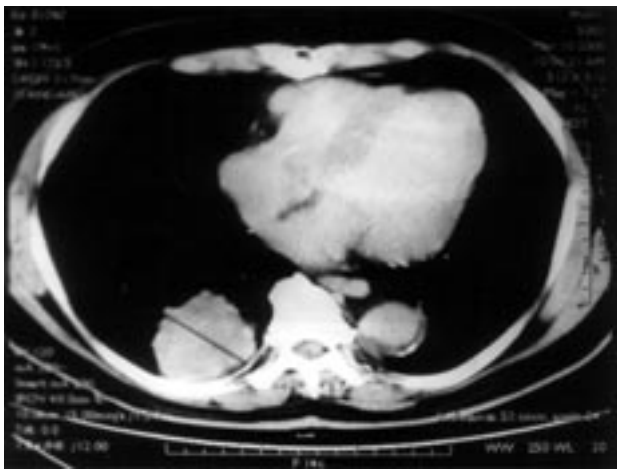

Fig 2. Thoracal CT shows multiple metastatic nodules in right hemithorax

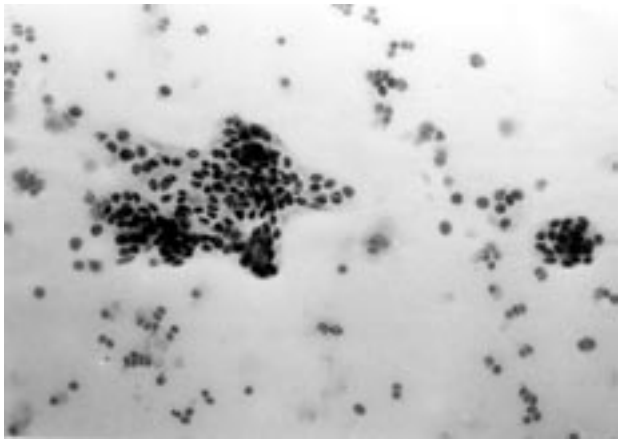

Fig 3. Smear of aspirate from the lung lesion shows monotonous tumor cells making spherical clusters with hyaline globules in the center of some clusters, consistent with metastatic adenoid cystic carcinoma (H&E, x200)

DISCUSSION

Adenoid cystic carcinoma which is frequently localized on the scalp is a rare tumor in external earway (4). This tumor usually grows slowly. Some factors including tumor positivity at surgical margins, parotid gland and adjacent bone involvement, perineural invasion and local recurrences are associated with aggressiveness and high mortality rate (5). When relapse develops, it generally occurs within two years. Late relapses were rarely reported as much as 14 years (5). In our case, pulmonary metastasis was detected 20 years after treatment, this is the longest period for metastases in the literature for adenoid cystic carcinoma. Rarely widespread visceral involvement may be seen (6). The performance status of patient and metastasis are important factors for selecting the treatment modality. The survival with surgery and postoperative radiotherapy is better than that with surgery alone (7). The degree of local invasiveness is important for planning surgical procedure. The modified temporal bone resection with radical excision of external earway has been reported as an effective treatment for these patients (4). Similar results were reported with step-wise surgery instead of en-bloc temporal bone and skull base resection (8). In tumors which were thought to be aggressive (invaded to the adjacent tissue, higher grade or impossible to get negative surgical margins) elective neck lymph node dissection could be added to surgery. Primary radical excision of tumor with lymph node dissection was performed in our case. The wide excisional surgery should be recom-

mended for the presence of invasion to the adjacent tissues (e.g. inner ear, temporal bone). Chemotherapy with known limited effect on metastatic adenoid cystic carcinoma patients, can be used for treatment. About 20% partial response was reported with weekly vinorelbine (30mg/m^2, iv) at salivary gland origin (9). In a patient with lung and liver metastasis, more than 50% regression was reported with docetaxel (100 mg/m2, iv, per 21 days) (10). Interestingly, one patient with tumor originated from parotid gland and relapsed after primary therapy (surgery + radiotherapy) a partial response was detected with tamoxifen alone (11). We treated our patient with gemcitabine + 5-fluorouracil regimen. After six cycles of chemotherapy, stable disease was seen on control CT imaging.

In conclusion, our case with adenoid cystic carcinoma is the one with pulmonary metastases in the longest period in English literature.

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