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SHORT REPORT

Adenoid Cystic Carcinoma of the External Auditory Canal With Pulmonary Metastasis

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Adenoid cystic carcinoma (ACC) of the external auditory canal (EAC) is a rare and specific variant of adenocarcinoma of the salivary gland (1). ACCs of the head and neck are usually found in the major salivary glands, as well as in the minor salivary glands such as the oral cavity, palate, nasal cavity, nasopharynx and lacrimal glands. ACCs of the external auditory canal arise from the ceruminous glands, sweat glands or ectopic salivary gland tissue (1). In general these tumors occur without specific symptoms or signs. ACCs may result in distant metastasis or recurrences (2).

Case report

A 58-year-old woman applied to the Otolaryngology Clinic with a 2-year history of purulent and bloody drainage from the right external ear canal. A previous biopsy at another center had revealed basal cell carcinoma 1.5 years before. We performed an exicisional biopsy. Gross and microscopic examinations of the lesion revealed a 0.9 x 1.1 cm diameter solid, white tumoral tissue obtained by excisional biopsy. Microscopic examination showed darkly stained cells with scant amphophilic cytoplasm and enlarged hyperchromatic nuclei. The cribriform and tubular components gave rise to a pathological diagnosis of ACC of the EAC. A computed tomography (CT) scan of the temporal bone demonstrated some slight thickening and mild contrast enhancement within the soft tissues of the right EAC (Figure 1). There was no bone involvement or regional lymph node metastasis. Cranial CT showed no intracranial invasion or metastasis. CT scanning of the thorax showed multiple pulmonary metastases (Figure 2). Although palliative surgery had been offered to the patient, she had declined the operation and chemotherapy was begun after the pathological diagnosis of ACC.

Malignant tumors of the EAC are rare and most are squamous cell carcinomas. ACC arising in the EAC is exceedingly rare. Although ACC is a rare EAC tumor, it is relatively common in the salivary glands of the head and neck (3). ACCs growth rate is slow and the nature of this carcinoma shows a slow malignant course (1). The main treatment is surgery. Postsurgical recurrences and metastasis to the lungs, regional lymph nodes and bones occur over many years. Slow growth and pulmonary metastasis were observed in our case and these findings were parallel to those in the literature (2-4,6). The true origin of ACC in the EAC is controversial. It has been proposed that these tumors arise from the ceruminous glands. Microscopic studies of these tumors and ceruminous glands demonstrate similar histologic features. Some authors have suggested that these tumors arise from the ectopic salivary glands of the EAC, although this opinion has not been proved (4). In general, ceruminous gland tumors can be classified as ceruminous adenoma, pleomorphic adenoma, ceruminous

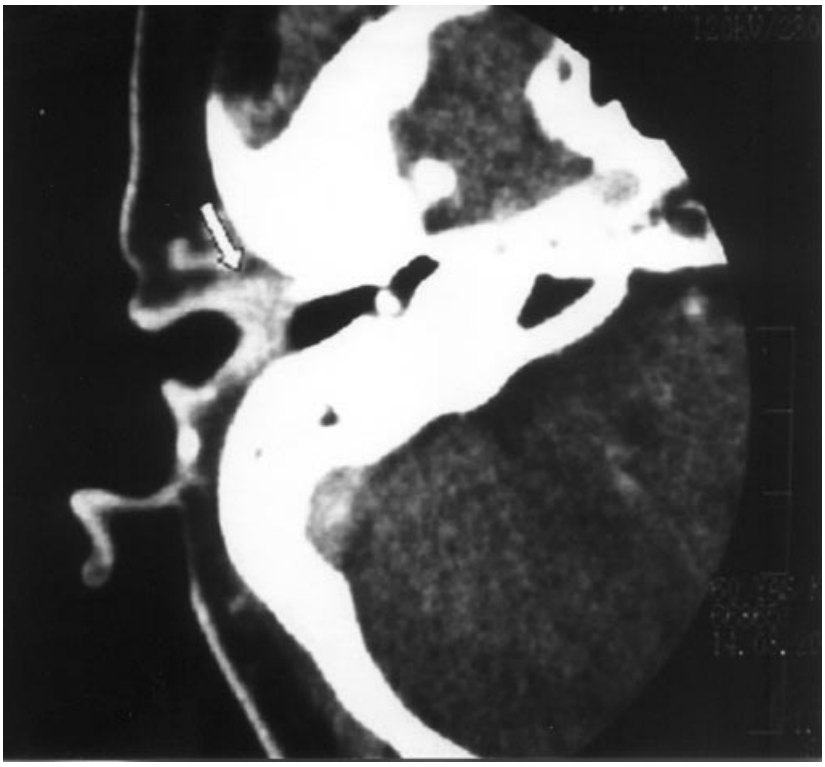

Figure 1. CT scan shows slight thickening and mild contrast enhancement in the soft tissue of the right external auditory canal.

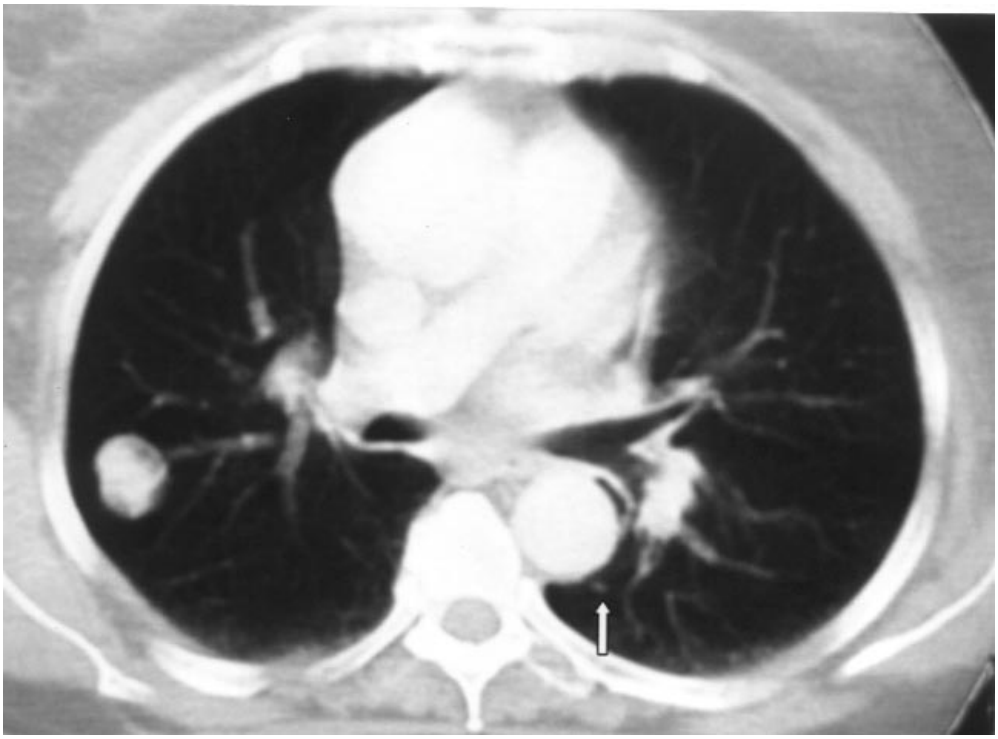

Figure 2. Thorax CT scan shows bilateral metastatic lesions in the lung.

adenocarcinoma and adenoid cystic carcinoma (3). ACC has 3 main histological patterns: tubular, cribriform and solid. In the salivary glands, the prognosis of ACC correlates with the predominant histological pattern. Tubular ACC has the best prognosis, whereas solid ACC has the worst prognosis (1). However, in the EAC no significant correlation between these histological patterns and prognosis has been demonstrated.

In our patient a bloody discharge from the right ear was the chief complaint. The clinical presentation of ACC of the EAC is variable although ear pain and hemorrhagic or purulent discharge are the most common complaints. Others symptoms include facial paresis, tinnitus and hearing loss. Our patient had undergone an aspiration biopsy that revealed basal cell carcinoma 1.5 years before admission to our hospital. There is no role for aspiration biopsy in such a lesion (5,7). Biopsy was delayed for more than 1 year, and at the time of diagnosis the patient had multiple pulmonary metastasis. Histological differential diagnosis of ACC should include basal cell carcinoma, ceruminous gland adenocarcinoma and ceruminous gland adenoma (2). ACC may locally invade soft tissue and bone, extend into the parotid gland and temporomandibular joint, invade by perineural and perivascular extension, and metastasize to the regional lymph nodes, lung and liver (5). Distant spread, most often to the lungs, and late recurrences are documented in the literature (6). Contralateral metastasis to the brain is also reported. Treatment consisted of radical excision of the EAC via a modified temporal bone resection (2). Radiotherapy and chemotherapy are not curative but can help in palliation and as adjuvant therapy.

ACC of the EAC is a rare tumor. ACC should be considered among the malignant tumors of the EAC. Lungs and regional lymph nodes must be evaluated with CT for metastasis.

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