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Adenoid cystic carcinoma of the parotid gland presenting as temporal bone neoplasm: a case report

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Abstract. *Adenoid cystic carcinoma of the parotid gland presenting as temporal bone neoplasm: a case report. Problem*: Temporal bone tumours are rare head and neck cancers that may involve both the parotid and the neck. In cases with concomitant temporal bone and parotid tissue involvement, tumour origin determination can be challenging. A tumour with parotid and temporal bone involvement is usually of temporal origin. The tumour may also be a rare parotid neoplasm with intratemporal invasion through the facial nerve, such as adenoid cystic carcinoma. These are slow-growing, yet locally aggressive tumours marked by late-stage distant metastases.
Case report: We describe a case of parotid adenoid cystic carcinoma invading the temporal bone via the facial nerve, presenting as primary temporal bone tumour with parotid tissue involvement. We emphasize the significance of intracranial involvement by perineural invasion.
Conclusion: Tumours mimicking temporal bone neoplasms can originate from the parotid tissue and invade through the facial nerve. Preoperative fine needle biopsy evaluation is indispensable for malignancy type determination and surgical planning.

Introduction

Temporal region tumours are very rare tumours that involve the temporal bone. They are classified into two groups based on origin: those originating from the temporal bone and those with local or metastatic involvement of the temporal bone.[1] Squamous cell carcinoma, basal-cell carcinoma, and parotid malignant neoplasm invading the temporal bone are the most frequent types.[1-3]

We present a patient with a temporal bone tumour, the diagnosis of which, based on pathological studies, was parotid adenoid cystic carcinoma (ACC) involving temporal bone. Although prognosis of extracranial perineural invasion is a well-known entity, there are relatively few cases of parotid ACC with intracranial extension via the facial nerve reported in the literature.

Case report

A 60-year-old female patient presented to our clinic with a one-year history of left peripheral facial paralysis and left otalgia and tinnitus gradually intensifying over the previous month. The patient did not complain of vertigo but did have a moderate degree of sensory-neural hearing loss in the ipsilateral ear. Physical examination revealed that the posterior wall of the external ear canal had protruded towards the lumen. The tympanic membrane was intact and dull. We palpated multiple lymphadenopathies along the jugular chain on the same side of the neck. House-Brackmann grade 4 peripheral facial paralysis was detected on the left side.

Computed tomography (CT) evaluation revealed a heterogeneous, hyper-dense, contrast-enhanced mass lesion occupying the rear wall of the left external ear canal, mastoid antrum, and tympanic cavity. The lesion was inflicting a focal destruction on the temporal bone in the neighborhood of the cochlea, with intracranial extension to the anterior petrous side, posterior temporal lobe, and cerebellar hemisphere, causing a defect inferiorly on the occipital bone and left lateral aspect of the first cervical vertebra (Figure 1). Inferomedially, the

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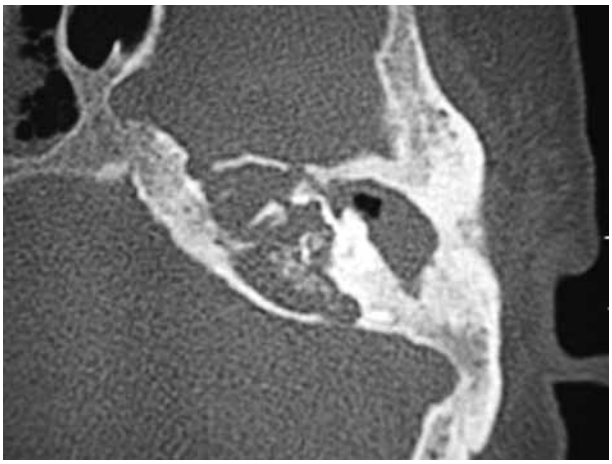

Figure 1
CT evaluation of the temporal region. A heterogeneous, hyperdense, contrast-enhanced mass lesion occupying the tympanic cavity and petrous side and extending through cerebral lobes.

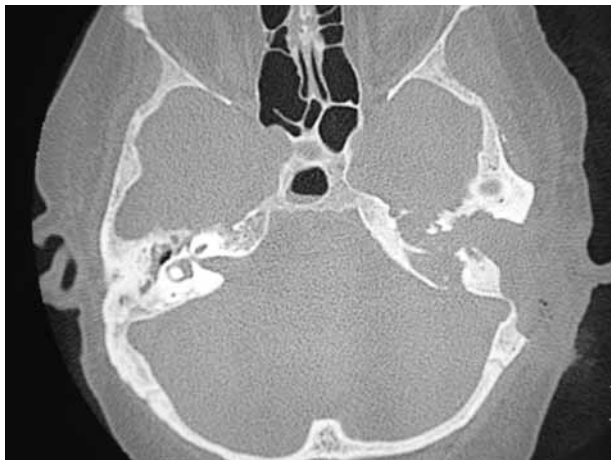

Figure 2
Equi-dense mass inferomedially continuous with the temporal mass through superficial and deep lobes of the parotid gland.

mass was destroying the sigmoid sinus and the mastoid part of the temporal bone with extension to the jugular foramen. On the inferolateral side, a continuity of the same mass through the parotid region is shown in Figure 2. Additionally, there was a mass of the same density present in the superficial and deep lobes of the parotid gland. Multiple lymphadenopathies were observed on the upper jugular chain. Magnetic resonance imaging (MRI) confirmed these findings.

These clinical and imaging findings suggested a primary temporal bone tumour, and thus, surgery was planned. Perioperatively obtained frozen sections from the middle ear indicated malignant epithelial tumour of salivary gland origin; subtotal left temporal bone resection and radical parotidectomy were performed. Perioperative evaluation of frozen sections of the upper jugular region lymphadenopathy identified malignant epithelial tumour metastasis, and left modified radical neck dissection was performed. During the operation, we observed that the tumour had invaded the area between geniculate ganglion and pes anserinus of the facial nerve. Since there was involvement of an extensive segment of the facial nerve and postoperative radiotherapy was required, we opted not to perform facial nerve grafting. Stylomastoid foramen and all of the mastoid apex cells were invaded by the tumour leaving no border between the parotid tissue and the temporal bone. The patient had postoperative grade V facial paralysis, but no other complications.

Pathological studies of the tissues from the parotid and temporal regions signified ACC, cervical lymphadenopathy at levels 2, 3, and 4, ACC metastasis, and severe perineural invasion by the tumour tissue. CT revealed a 1 cm diameter residual mass on the occipital basis and around the hypoglossal and carotid canals.

The patient then received radiotherapy due to the presence of neck metastasis, aggressive local invasion features, and a residual mass. Five thousand cGy were delivered to the primary site, an additional 1000 cGy were delivered to the neck, and 1000 cGy to the temporal region. Tarsoraphy was performed in order to protect the eye. Sixth months postoperatively there was no additional mass at either the parotid or the neck region, although there was grade-five facial paralysis and the same sized residual mass was present on CT.

Discussion

Secondary malignancies of the temporal bone are rare[2] and it would be unusual to suspect secondary involvement of this region by the tumour prior to pathological diagnosis. The patient presented in this study was diagnosed with a temporal bone neoplasm with extracranial local invasion, and underwent surgery. However, surgical and pathological evaluations showed parotid ACC invading the temporal bone through the facial nerve. Tumours

that invade via the facial nerve are most frequently neuromas of the facial nerve itself or extraneural facial hemangiomas,[4] both of which are benign. Invasion of the facial nerve by extratemporal malignant tumours along the intratemporal route is rare, and usually progresses with extratemporal involvement. There are, however, very rare cases of intratemporal and intracranial invasion through the facial nerve.[5-7] Perineural invasion is characteristic of parotid ACC. Facial paralysis is present in 15% of patients with parotid carcinomas, and in 70% of patients with parotid ACCs.[5,7-9] In this case, parotid ACC extended along facial nerve trace, leading to invasion of temporal cells. The prognosis of the tumour is determined by neural involvement, which is based on various mechanisms: direct invasion throughout perineural space, compression, or extension leading to nerve paralysis and invasion.[6,10]

In secondary malignancies of the temporal bone, loss of hearing and vertigo are the most frequent symptoms. Other presenting symptoms may be discharge from the external ear meatus, otalgia, and other cranial nerve involvement findings including isolated facial nerve paralysis. Patients may remain asymptomatic for a long time.[6] In this case, the one-year history of facial paralysis is highly characteristic of parotid neoplasm; however, accompanying otalgia and the absence of marked palpable parotid mass suggested a temporal bone tumour rather than parotid neoplasm. With primary parotid malignancies, we expect the salivary gland contours to be highly damaged, whereas with secondary parotid invasions of temporal bone tumours the parotid gland contours are more regular. Similarly, in primary temporal bone tumours, destruction of the external ear canal, middle ear, and mastoid cells are more frequent than in secondary bone involvement of parotid neoplasm.[2]

In such cases, the history and physical examination do not provide sufficient evidence, and CT and MR images should be scrutinized carefully in order to obtain sufficient information about the nature of the tumour, based on the areas involved, and the extent of invasion for preoperative planning. Stylomastoid foramen or mastoid-type bone erosion or enlargement of the facial canal within the temporal bone on CT are suggestive of parotid neoplasm perineurally invading the temporal bone.[2,11]

Here, the patient had severe local destruction of the rear wall of the external ear canal, mastoid antrum, and mastoid cells of the medial section. These findings should have provoked suspicion of a primary parotid malignancy, but the absence of massive involvement of the gland on radiological evaluation diverted this suspicion. The treatment for both primary temporal bone tumour and temporal bone involvement secondary to parotid carcinoma is parotidectomy, subtotal petrosectomy, neck dissection, and postoperative radiotherapy. The only difference in treatment strategies is the approach to the facial nerve. With temporal bone tumours, the facial nerve is protected while superficial or deep parotidectomy is performed. With temporal bone involvement secondary to parotid ACC, the facial nerve is sacrificed by radical parotidectomy due to severe perineural invasion. In this case, intraoperative pathological findings led us to sacrifice the nerve.

Although intraoperative frozen section evaluation provided us accurate information for correct surgical management, the preferred approach would have been preoperative evaluation of fine needle aspiration biopsy from both the gland and the cervical node. The presence of an intact tympanic membrane and the absence of a mass either behind the eardrum or at the parotid gland directed us to make an intraoperative pathological evaluation. However, in cases with facial paralysis and expansive mastoid cell involvement, primary suspicion should be directed at parotid malignancy, and preoperative cytological evaluation should be performed. In tumours with parotid tissue and temporal bone involvement, tumour origin determination is challenging. History and physical examination findings are helpful; however, the presence of focal bone erosion on the facial nerve trace on CT suggests a neoplasm of parotid origin with temporal bone involvement, and the diagnosis should be confirmed by preoperative fine needle biopsy.

Conclusion

Tumour origin determination is highly challenging in cases with concomitant temporal bone and parotid tissue involvement. Tumours mimicking temporal bone neoplasms can originate from the parotid tissue and invade through the facial nerve. Preoperative fine needle biopsy evaluation is indispensable for malignancy type determination and surgical planning.

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