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Case Report Open Access

Imaging of Auriculotemporal Nerve Perineural Spread

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Abstract

Importance: Adenoid cystic carcinomas are relatively rare tumors, notorious for wide local infiltration and perineural spread. Perineural extension commonly occurs along branches of the trigeminal and facial nerves, and its presence represents a poor prognostic factor with implications upon treatment approach.

Observations: We report a case of a 61-years-old female presenting with worsening left facial numbness and weakness. On magnetic resonance imaging, the patient was found to have perineural spread of a left parotid tumor along the auriculotemporal nerve. There was involvement of the V2 and V3 branches of the trigeminal nerve. An ultrasound-guided biopsy of the mass demonstrated adenoid cystic carcinoma.

Conclusions and relevance: The auriculotemporal nerve may serve as a route for tumor spread, particularly in the setting of head and neck malignancy. Moreover, this particular suspicion should be raised when patients with known malignancy experience concomitant trigeminal (V) and facial (VII) nerve dysfunction.

Keywords: Perineural spread; Head and neck malignancy; MRI; CT

Introduction

Adenoid Cystic Carcinoma (ACC) is a slow-growing, but aggressive malignant tumor with a propensity for local recurrence and late distant metastasis. Although it is relatively rare, constituting only 1% of all malignant oral and maxillofacial tumors, ACC comprises approximately 15% of parotid gland malignancies [1,2]. It is characterized by wide local infiltration and is well known for its tendency for perineural spread. Epidemiologically, ACC exhibits a slight female predilection, and has a peak incidence in the 5th and 6th decades of life [2]. Clinically, patients typically present with signs and symptoms related to local tissue invasion and perineural spread [3]. The most commonly involved nerves are the facial nerve, as well as the maxillary (V2) and mandibular (V3) divisions of the trigeminal nerve [4,5]. It is thought that pre-existing connections between the facial and trigeminal nerve, including the auriculotemporal nerve, aid in the perineural dissemination of tumor between these two nerves. In this report, we present a case of ACC arising in the parotid gland with extension along the auriculotemporal nerve.

Case Report

A 61 years-old woman presented 4 years prior with a painless mass involving the left parotid region. The patient's past medical history was non-contributory. There were no other neurologic signs or symptoms, and no history of malignancy. A CT scan of her head performed 4 years previous demonstrated an ill-defined lesion in the superficial left parotid gland that corresponded to a palpable preauricularmass (Figure 1). The clinician recommended that this lesion be followed conservatively with observation. However, the lesion gradually increased in size over time. The patient then developed increasing numbness in the left mandibular region, weakness of the ipsilateralfrontalis muscle and pain and left-sided trismus. An ultrasound-guided core biopsy was performed which demonstrated an intermediate grade (2/3) adenoid cystic carcinoma. She was then referred to our tertiary oncology center. Physical examination revealed a firm, tender left parotid mass as well as left Level II adenopathy. There was numbness involving the left V2 and V3 distribution and slight left facial nerve weakness.

A Magnetic Resonance Imaging (MRI) examination was performed (Figure 2). This demonstrated a lobulated mass measuring 2.0×2.2 cm within the superficial lobe of the left parotid gland with extension to the overlying capsule and thickening of the overlying skin. Thick curvilinear enhancing tissue was noted to be extending from the parotid mass around the posterior ramus of the mandible and joining with the V3 trunk in the left masticator space which corresponds to the track of the auriculotemporal nerve. Contiguous thickening and enhancement along V3 superiorly through a widened foramen ovale

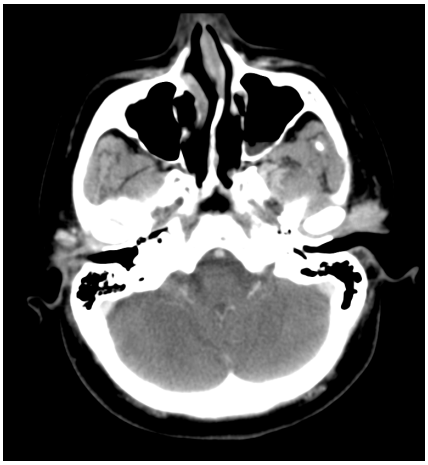

Figure 1: Contrast enhanced axial CT image shows an enhancing, lobulated mass in the left preauricular region.

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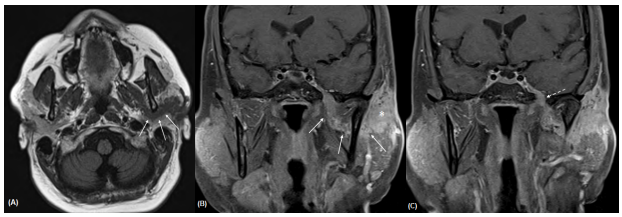

Figure 2: Axial T1 weighted image (A) and post contrast coronal T1 weighted images with fat saturation (B, C) demonstrates a curvilinear band of enhancing tumor (arrows in A, B) that extends from the left parotid mass (asterisk) and tracks behind the mandibular ramus and joins with the V3 nerve in the masticator space. There is contiguous extension along V3 superiorly through a widened foramen ovale (dashed arrow in C) and asymmetric thickening and enhancement of the left cavernous sinus.

was noted. There was also slight thickening in the adjacent inferior aspect of the left cavernous sinus. These findings were compatible with perineural tumor spread along the left auriculotemporal nerve with contiguous extension to involve the left V3.

The patient underwent left radical parotidectomy and neck dissection with sacrifice of the left facial nerve. Visible perineural tumor spread along the ATN was noted during surgery. A composite resection of the left mandible and dissection into the left infratemporal fossa was also performed. A radial forearm flap was used to reconstruct the soft tissue defect. A sural nerve graft was taken to reconstruct the left facial nerve which was connected to the ipsilateral hypoglossal nerve. The patient then underwent a course of radiation therapy, completing a course of 66Gy in 33 fractions.

Discussion

ACC is one of the most common salivary gland malignancies. As illustrated in our patient, this is a locally invasive tumor with a notorious tendency toward perineural spread. In this case, the tumor was found to extend along the left auriculotemporal nerve with involvement of the mandibular (V3) division of the left trigeminal nerve.

The auriculotemporal nerve is one of the major known communications between the facial and trigeminal nerves. It arises as two nerve roots from V3 that course posteroinferiorly around the middle meningeal artery–the upper and lower roots extend lateral and medial to the artery, respectively– and coalesce into a short trunk medial to the Temporomandibular Joint (TMJ) and superior to the bifurcation of the external carotid artery into its temporal and maxillary branches. From there, it divides into multiple branches, including the anterior and posterior communicating rami that join the facial nerve within the parotid gland to supply sensory fibers to its zygomatic, buccal, and mandibular divisions [5]. Although not present in our patient, symptoms suggestive of auriculotemporal nerve involvement include periauricular pain and TMJ dysfunction and/or tenderness [5].

Imaging features of perineural tumor spread include thickening and abnormal enhancement along involved nerves, foraminal widening and erosions due to tumor growth along the nerves, neuropathic atrophy of denervated muscles, as well as obliteration of perineural fat pads [4,6]. For optimal detection of perineural invasion, high-resolution, fat-suppressed axial and coronal T1-weighted MR imaging with and without contrast is recommended [4-7]. Lesions typically exhibit abnormal contrast enhancement on post-contrast images, as well as abnormal signal hyperintensity on T2-weighted images [4]. However, negative radiologic studies may be obtained even in patients with tumor invasion [4,6]. Therefore it is important to correlate radiologic findings with clinical and pathologic data [8].

Histologically, ACC is composed of a mixture of epithelial and ductal cells, and is assigned one of three histologic grades: Grade I, a well-differentiated tumor composed of tubular and cribriform patterns without solid components; Grade II, a tumor with a pure cribriform or mixture of patterns with solid growth pattern less than 30% of the tumor; and Grade III, a tumor with marked predominance of the solid pattern [9]. Higher grades are associated with larger size at presentation, higher recurrence rates, and higher mortality rates. Other prognostic factors include tumor location, stage, presence or absence of surgical margins, and the anatomic structures involved (i.e. perineural spread) [2,9].

Our patient underwent surgical resection of the mass with radical parotidectomy followed by adjuvant radiotherapy. Treatment of ACC is primarily surgical with or without radiation therapy. The goal of surgery is to obtain at least a 1-cm margin around the tumor [2]. However, the disease is extremely difficult to treat due to high rates of recurrence and metastases if the patient lives long enough even despite radical surgery. The five-years survival rate after effective treatment is 75%, while long term prognosis is poor with a 10 years survival rate of 20% and 15 years survival rate of 10% [2]. Adjuvant radiotherapy combined with radical surgery has been shown to increase long-term survival.

The presence of perineural invasion represents a poor prognostic factor and has implications on the treatment approach, such as indicating the need for wider resection and expanded radiation field [4,10]. Tumors with perineural spread have high recurrence rates and decreased survival. Patterns of perineural spread and knowledge of the anatomy of the cranial nerves may help in planning the appropriate treatment modalities.

As we demonstrate, the auriculotemporal nerve can serve as a route for tumor spread in the setting of head and neck malignancy. Moreover, this particular suspicion should be raised when patients with known malignancy experience concomitant trigeminal and facial nerve dysfunction.

We report a case of an adenoid cystic carcinoma of the parotid gland with perineural invasion of the auriculotemporal nerve and involvement of V2 and V3. High-resolution MRI with and without contrast is the imaging modality of choice for detection of perineural tumor invasion. Due to its implications on prognosis and treatment, it is imperative to recognize the presence of perineural invasion.

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