

## Case Report

# Extensive Resection of Invasive Recurrent Left Parotid Gland Carcinoma ex Pleomorphic Adenoma

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A 50-year-old man experienced tumor recurrence with invasion into the skull base. Preoperative neuroimaging showed partial destruction of the body of the mandible and the anterior and middle cranial bases. Further, there was tumor involvement of the left internal carotid artery. Preoperative embolization of the left internal carotid artery and an extensive resection of the tumor were successfully performed. Even though salivary gland tumors are rarely treated by neurosurgeons, a multidisciplinary approach allowed effective treatment of this tumor. The optimal therapeutic strategy for this rare type of intracranial invasion is described.

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## Introduction

Neurosurgeons are not typically employed for the treatment of salivary gland tumors, including adenomas, carcinoma ex pleomorphic adenoma, or adenoid cystic carcinomas (cylindroma).<sup>1</sup> However, rare cases of intracranial invasion by such tumors have been reported<sup>2-8</sup> that require management by a neurosurgeon. In the case of intracranial invasion, the tumor infiltrates via the foramina of the cranial base and usually demonstrates perineural spread.<sup>2</sup> There have been tremendous advances in surgical modalities for cranial base surgery, and interdisciplinary collaboration is often required to achieve curative treatment. The present study describes a case of recurrent pleomorphic adenoma ex carcinoma with invasion into the skull base. This tumor was successfully removed using a combined strategy consisting of endovascular, neurologic, and plastic surgeries.

## Case Report

A 50-year-old man underwent partial removal of a left parotid salivary gland pleomorphic adenoma in 1984. The patient underwent a second operation in 1987 and a third operation in 1989 for tumor

regrowth. Tumor regrowth was noted again, and the patient underwent focal irradiation (65 Gy) in 1995. Despite these procedures, the patient experienced progressive left cranial nerve palsies involving the Vth to XIIth nerves, and outward protrusion of the tumor resulted in cosmetic problems. Thus, the patient was referred to our department for further evaluation and treatment.

On admission, a left sub-auricular elastic hard tumor of 4 cm in diameter was noted. Neurological examination revealed left peripheral facial palsy and Villaret syndrome. Computed tomography (CT) demonstrated a multi-cystic tumor of 8 cm in diameter with capsular enhancement located in the left infratemporal region. A bone imaging CT demonstrated destruction of the temporal, occipital, and mandible bones. Magnetic resonance image (MRI) demonstrated a high intensity mass on the T2-weighted image, and Gd-DTPA enhancement showed compression of the left internal carotid artery (ICA) towards the right without any cavernous sinus invasion (Figure 1). The tumor also invaded into the posterior fossa via the left jugular foramen and involved the left vertebral artery.

A cerebral angiogram (CAG) revealed wall irregularity in the left ICA at the petrous portion. Balloon test occlusion (BTO) of the left ICA was well tolerated, and coil embolization of the left ICA was performed before surgery without any sequelae (Figure 2). Total

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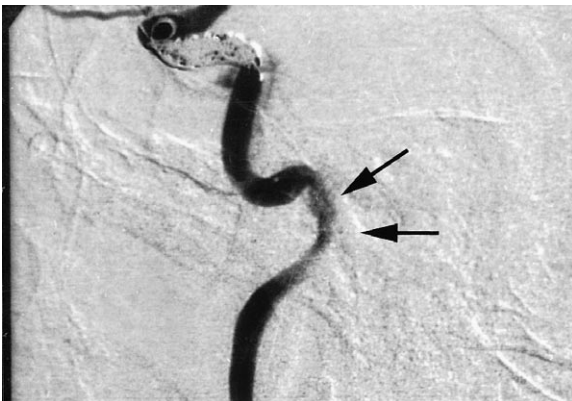
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**Figure 1.** Preoperative axial T1-weighted magnetic resonance (MR) image with contrast medium reveals a well-enhanced tumor with invasion into the deep skull base structures (arrowheads). The arrow indicates the left internal carotid artery.



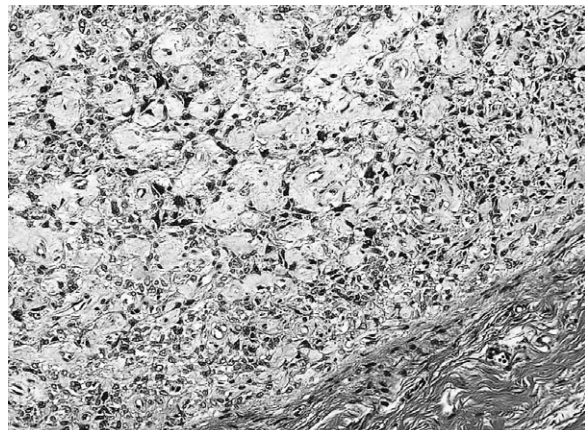
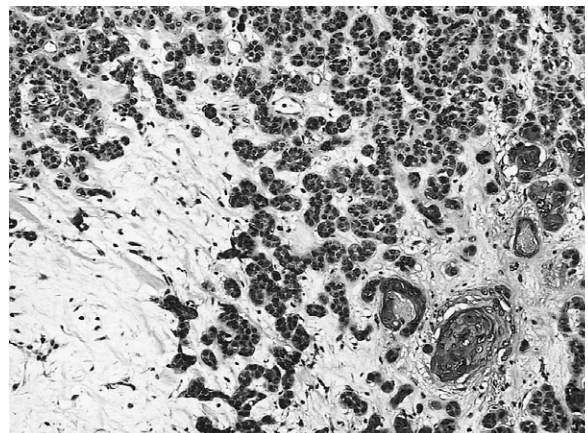
**Figure 2.** Lateral view of the left internal carotid artery angiogram on coil and balloon embolization. The petrous portion is narrowed by tumor (arrows).

tumor removal was performed one month after the coil embolization. The surgery was performed as follows: (1) demarcation from the left subauricular tissue; (2) a left frontotemporoccipital zygomatic approach; (3) a left mandibulectomy and tumor removal from the infratemporal fossa; (4) sacrifice of the left ICA and the jugular vein; (5) removal of extradural tumor invasion into the posterior fossa; and (6) plastic repair of the tissue defect using a pediculated musculocutaneous flap of the *latissimus dorsi* on the left back side through the subcutaneous tunnel (Figure 3). All procedures were performed extradurally.

Histological examination showed that the tumor consisted of a myxomatous stroma, tubular epithelium, and cartilage formation.



**Figure 3.** Postoperative image of the musculocutaneous flap on the operative field.



**Figure 4.** Photomicrograph of the surgical specimen. **A.** Main tumor. The tumor consists of small duct formations and shows squamous differentiation. The stroma has myxoid matrix simulating cartilaginous differentiation (HE stain,  $\times 130$ ). **B.** Dermal invasive lesion. The proliferating epithelial cells show marked cellular atypia, indicating malignant change of pleomorphic adenoma. Dermis is observed in the right lower corner (HE stain,  $\times 130$ ).

Dermal invasion with a few atypical cells was detected. The final diagnosis was carcinoma ex pleomorphic adenoma (Figure 4).

Total removal of tumor was confirmed by postoperative CT and MRI. No postoperative adjuvant therapy was performed. The patient has not experienced tumor recurrence over the 2-year follow-up period.

## Discussion

Most salivary gland tumors consist of benign mixed tumors, Carcinoma ex pleomorphic adenoma only constitutes 1.5-12% of all cases<sup>1-4,9</sup> and typically affects patients in their sixth to eighth decades of life, with a slight female sex preponderance.<sup>9</sup> This tumor is thought to result from a malignant transformation of a previous pleomorphic adenoma, and it contains only malignant epithelial component. In contrast, malignant mixed adenoma has both mesenchymal and epithelial malignant components.<sup>1,10</sup> The single most reliable prognostic marker for patients with carcinoma ex pleomorphic adenoma is the extent of tumor infiltration beyond the capsule and intracapsular type of tumor, which neither recurs nor metastasizes.<sup>9</sup> Other important prognostic factors are tumor size, histological grading of the malignant component, and degree of surgical resection.<sup>9</sup> Several studies have reported that one-third of incompletely resected benign tumors will eventually transform into malignant tumors, typically with the 4th recurrence at an average of 15.8 years.<sup>3,11</sup> Of the malignant parotid tumors, adenoid cystic carcinoma has a greater tendency to metastasize than any other type of tumor.<sup>3</sup>

The frequency of intracranial invasion of adenoid cystic carcinoma ranges from 4-22%.<sup>4,12</sup> This occurs via tumor invasion through the nerve sheath of the mandibular and maxillary nerves, Eustachian tube, ICA, and/or via a hematogenous route.<sup>2,4,5,8</sup> The tumor also invades the gasserian ganglion via the Eustachian tube.<sup>6</sup> The fifth cranial nerve is most frequently affected cranial nerve in the context of intracranial invasion.<sup>2,6</sup>

When carcinoma ex pleomorphic adenoma recurs or metastasizes, it usually spreads via lymphatics or directly invades surrounding neurons and vessels, but hematogenous dissemination rarely occurs.<sup>13</sup> Perineural spread is thought to be an aggressive manifestation despite its infrequency in the case of skin cancer.<sup>14</sup> Further, this type of invasion may be an indication for aggressiveness even in benign salivary gland tumors.

Other reported routes into the intracranial region include the cribriform plate, paranasal sinuses, and the petrous apex.<sup>2,4,8</sup> In this case, the tumor invaded via the foramen lacerum, the jugular foramen, and the hypoglossal canal, which resulted in cranial nerve deficits. Although the presence of facial nerve involvement typically indicates either long-term persistence or noncurability of the adenocystic carcinoma, we could not conclude whether the facial nerve involvement in the present case indicated a favorable or poor prognosis.<sup>15</sup>

Adenoid cystic carcinoma tends to invade perivascularly, which is consistent with the invasion pattern and ICA involvement in the present case.<sup>12</sup> The management of the ICA is a very important consideration for surgical treatment, and Gromley reported that 13 of 16

patients showed tumor involvement of the ICA requiring sacrifice of the ICA in 3 of 5 patients with severe encasement and narrowing.<sup>2</sup>

Published data regarding neuroradiologic findings of intracranial salivary gland tumors, particularly carcinoma ex pleomorphic adenoma, are rare. Ossification indicates that the origin of the tumor was likely a benign pleomorphic adenoma.<sup>16</sup> Further, the tumor typically contains some necrotic tissue and is homogeneously enhanced by contrast medium on CT and MRI. Dural enhancement has been detected in some cases, which reflects tumor invasion into the dura mater.<sup>4</sup> In the present case, no sign of tumor calcification was noted, and there was marginal enhancement on CT and homogeneous enhancement on MRI without a dural tail sign.

A radical neck resection is not indicated as part of initial therapy and re-operation for simple recurrence. However, radical resection should be performed when cervical neck lymph nodes or other structures are involved.<sup>15</sup> Gormley et al.<sup>3</sup> reported a mean survival of 49 months (range 6-98 months) after radical surgery, which supports the utility of radical surgery. Total parotidectomy or subtotal petrosectomy with facial nerve resection may also reduce the recurrence rate of pleomorphic adenoma.<sup>11,17</sup> Therefore, tumors with malignant components should be resected along with the affected facial nerve, as in the present case.

Shotton et al.<sup>5</sup> also reported that permanent balloon occlusion of the ICA improved access to the deep-seated structures. The patient in the present report tolerated BTO well, and coil embolization of the left ICA was performed. In patients unable to tolerate BTO, the ICA may have to be sacrificed using extracranial-intracranial artery bypass in order to perform a total removal of the tumor.<sup>5</sup> After a radical resection of the middle and posterior cranial fossa, the temporalis muscle flap is typically used for repairing surgical defects.<sup>5</sup> However, in our patient, the surgical defect was too large to repair in this manner, and the tumor had tightly adhered to the skin. We therefore used a musculocutaneous flap from the *latissimus dorsi*.

Most postoperative complications are related to cranial nerve injury and cosmetic problems.<sup>5</sup> Further cranial nerve transplantation or plastic surgery may be considered to optimize patient functional status or aesthetics.

The use of postoperative adjuvant therapy remains controversial. In general, chemotherapy for this slow growing tumor is ineffective, and irradiation is the treatment of choice for tumor control<sup>1,2,6,8</sup> but is unlikely to induce complete remission.<sup>2</sup> Regardless, except for carcinomatous components, irradiation appears indicated.<sup>12,15</sup>

In conclusion, invasive salivary gland tumors require complete resection with the assistance of neuroradiological techniques rather than partial tumor removal combined with adjuvant chemotherapy and/or radiotherapy.

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