

# Intrasellar Remote Metastasis from Adenoid Cystic Carcinoma of Parotid Gland: Case Report

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**Abstract.** Adenoid cystic carcinoma is a tumor of exocrine glands originating primarily from the minor and major salivary glands, lacrimal gland, bronchus, breast, and intestinal and genital tracts. Intracranial remote metastasis from adenoid cystic carcinoma in salivary gland is quite rare. The authors encountered a case of intrasellar remote metastasis from an adenoid cystic carcinoma of parotid gland origin, presenting with hyponatremia secondary to the syndrome of inappropriate secretion of antidiuretic hormone (SIADH). To our knowledge, this is the first reported case of metastasis from an adenoid cystic carcinoma to intrasellar area. A 78-year-old woman had an adenoid cystic carcinoma in the left parotid gland, which was resected surgically followed by local radiation therapy of 60 Gy. After 4 years, the patient presented with general malaise, followed by disturbed consciousness caused by hyponatremia. The clinical data showed severe hyponatremia induced by SIADH. An intrasellar heterogenous mass lesion compressing the optic chiasm was resected subtotally via an endonasal transsphenoidal approach. Histopathological examination of the tumor specimens revealed adenoid cystic carcinoma, which had identical histological findings as those of the painful superficial cervical lymph nodes resected in the same operation. Tumors such as the present case are easily confused with pituitary adenoma or craniopharyngioma. Although rare, metastasis from tumors including those of salivary gland origin should be considered in the differential diagnosis of unusual pituitary tumors.

**Key words:** Pituitary gland, Adenoid cystic carcinoma, Metastasis, Salivary gland, SIADH

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**ADENOID** cystic carcinoma is a tumor of exocrine glands originating primarily from the minor salivary glands of upper respiratory gland, the major salivary glands, lacrimal gland, bronchus, breast, and intestinal and genital tracts [1, 2]. It is characteristically slow growing, but often invades the cranial base directly with rates of cranial base invasion between 4 and 22% [3]. However, intracranial remote metastasis from adenoid cystic carcinoma of the salivary gland is quite rare [4].

We encountered a case of intrasellar remote metastasis from an adenoid cystic carcinoma of parotid gland origin, presenting with hyponatremia secondary to the syndrome of inappropriate secretion of antidiuretic

hormone (SIADH). To our knowledge, this is the first reported case of remote metastasis from an adenoid cystic carcinoma in parotid gland to intrasellar area.

## Case Report

### History

A 78-year-old woman had an adenoid cystic carcinoma in the left parotid gland, which was resected surgically followed by local radiation therapy of 60 Gy. In the same year, the tumor recurred locally and tumor removal and segmental resection of the mandibular bone with reconstruction of mandible and middle pharynx were performed. Radiation therapy of 60 Gy was additionally delivered to the skull base. Since then, the patient had been stable without recurrence for 4 years. Then she presented with general malaise, followed by disturbance of consciousness with hyponatremia.

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## Examinations

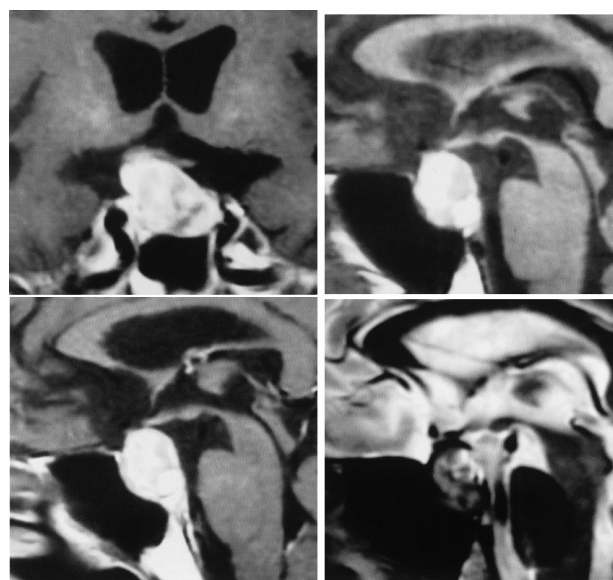
When the patient was admitted to the former hospital, laboratory analysis revealed the following; serum sodium (Na): 104 mEq/l, potassium: 4.1 mEq/l, plasma osmolality (Posm): 206 mOsm/kg, blood urea nitrogen (BUN): 10.2 mg/dl, serum creatinine: 0.5 mg/dl, plasma renin activity (PRA): 0.3 ng/ml/h, aldosterone: 82 pg/ml, atrial natriuretic peptide (ANP): 5 pg/ml, total protein: 6.5 g/dl, total cholesterol: 202 mg/dl, urine Na: 124 mEq/day, and urinary osmolality (Uosm): 366 mOsm/kg. Endocrinological tests (serum concentrations) disclosed the following; ACTH: 15 pg/ml, cortisol: 25.9 µg/dl, TSH: 0.727 µIU/ml, FT4: 0.6 ng/dl, PRL: 26.2 ng/ml, and AVP: 1.2 pg/ml despite the marked hypoosmolality. The anterior lobe function of the pituitary gland remained normal. The patient was not thirsty, and had neither dry skin or tongue, nor pretibial edema. The serum Na was normalized 10 days after starting treatment of water intake restriction and induced Na replacement. Hyponatremia did not recur thereafter at all. Posm also increased to the normal range 10 days after admission and remained at normal level. Plasma AVP levels had been within normal limit between 1.1 and 1.8 pg/ml during the all progress. These clinical and laboratory features were consistent with the diagnostic criteria of SIADH.

Magnetic resonance (MR) images demonstrated an intrasellar mass with suspected intratumoral hemorrhage.

Two months later, the patient was transferred to our institute for further examinations and treatment for the intrasellar mass lesion. Serum Na, Posm, and plasma AVP were normal. The patient had malaise, emaciation, and anorexia. Swelling of left superficial cervical lymph nodes was recognized. A computed tomography (CT) scan showed a heterogenous slightly hyperdense lesion. A MR imaging study demonstrated a heterogenous intrasellar mass lesion compressing the optic chiasm with partial gadolinium enhancement (Fig. 1). Cerebral angiography revealed a mass without definite abnormal vascularity. Based on these preoperative studies, the tumor was considered to be a metastasis with intratumoral hemorrhage, a craniopharyngioma, or a pituitary adenoma with apoplexy.

## Operation

The patient underwent an endoscope-assisted tumor



**Fig. 1.** Preoperative T1-weighted magnetic resonance images (upper left; coronal without enhancement, upper right; sagittal without enhancement, lower left; sagittal gadolinium-enhanced) and T2-weighted magnetic resonance sagittal image (lower right) demonstrating a heterogenous intrasellar lesion suspected to have intratumoral hemorrhage, with suprasellar extension compressing the optic chiasm showing partial gadolinium enhancement.

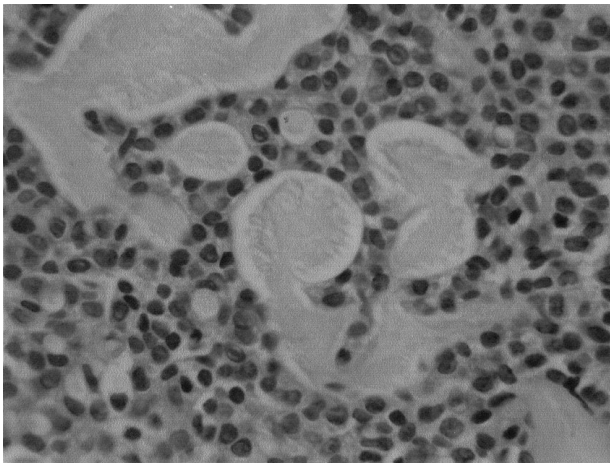
resection via an endonasal transsphenoidal approach for the tumor [5–7]. After a crosswise dural incision, the old intratumoral hematoma was discharged. The tumor was not clearly separated from the normal pituitary gland and was therefore resected subtotally. Prior to the endonasal resection of the tumor, we extirpated the painful superficial cervical lymph nodes on the left side, suspected to be metastasis from the parotid gland adenoid cystic carcinoma.

## Histological findings

Histopathological examination of the tumor specimens revealed adenoid cystic carcinoma with a MIB-1 index of 12.5% (Fig. 2), which had identical histological findings as those of the superficial cervical lymph nodes resected in the same operation.

## Postoperative course

The postoperative course was uneventful and the patient did not develop any additional pituitary hypo-



**Fig. 2.** A photomicrograph of the tumor specimens obtained during surgery showing a typical cribriform pattern with mucinous materials, which are typical histological features of adenoid cystic carcinoma (hematoxylin & eosin stain, original magnification  $\times 200$ ). The finding was identical to the histology of the superficial cervical lymph nodes resected in the same operation.

functions following the surgery. Hyponatremia has also not recurred since the surgery. Furthermore, Posm and plasma AVP have been normal postoperatively. The patient is being followed as an outpatient and has been scheduled to receive gamma knife radiosurgery for the small residual intrasellar tumor.

## Discussion

### *The features of adenoid cystic carcinoma and salivary gland tumors of the sellar region*

Adenoid cystic carcinomas are characterized by slow growth but peritumoral invasion or perineural infiltration. Early local recurrence and late distal metastasis are common in spite of radical extirpation of the primary tumor [4, 8, 9]. According to Shotton *et al.* [10], cranial base invasion occurs along three routes: the eustachian tube, the mandibular and maxillary nerves, and the internal carotid artery. However, intracranial remote metastasis from adenoid cystic carcinoma of salivary gland origin is quite rare [4]. In the present case, metastasis from an adenoid cystic carcinoma to the intrasellar region was diagnosed histologically.

There have been some reported cases of salivary gland-like tumors originated from the sellar and para-

sellar regions [17–19]. The presumed origin of these rare neoplasms is embryological remnants of the salivary gland within the normal pituitary gland.

### *Pituitary metastasis*

Pituitary gland is a common target organ of metastatic tumors despite its small volume. The incidence of pituitary metastases has been variously reported to be from 1 to 26.6% in autopsy [11–13]. Breast carcinoma has an unusually high incidence of pituitary metastasis, ranging from 9 to 29% [12, 14]. There are 4 metastatic pathways to the pituitary gland; 1) direct blood-borne metastasis to the posterior lobe with subsequent expansion, 2) blood-borne metastasis to the pituitary stalk with growth into the anterior and posterior pituitary lobes, 3) blood-borne metastasis to the clivus, dorsum sellae, or cavernous sinus, which then spreads into the pituitary gland, and 4) leptomeningeal spread with involvement of the pituitary capsule [12, 15, 16]. Of these 4 routes, the incidence of direct blood-borne metastasis to the posterior lobe with subsequent expansion may be the most highest. For the location of metastases, the posterior lobe alone was the most common, ranging from 50% to 52% of pituitary metastases. On the contrary, metastasis to the anterior lobe alone occurs in 21% [15].

In the present case, SIADH was improved with treatment of water intake restriction and induced Na replacement and did not recur even before adenomectomy. In case of direct metastasis to the posterior lobe, SIADH must be more persistent and recurrent even after treatment.

### *Symptoms associated with pituitary tumors*

Diabetes insipidus (DI) is the most common symptom of metastatic pituitary tumors. DI occurs in 33 to 70% of the patients with symptomatic pituitary metastases; in contrast, it occurs in only 1 to 2% of patients with pituitary adenomas [12, 15, 20, 21]. According to the data of 36 cases of symptomatic pituitary metastasis reported by Morita *et al.*, DI was seen in 61%, anterior pituitary insufficiency in 47%, and retro-orbital pain in 39% [12].

There are no reports of metastatic pituitary tumor cases manifesting SIADH. Pituitary adenomas and craniopharyngiomas very rarely present with SIADH [22–25]. The present case of pituitary metastasis

from an adenoid cystic carcinoma of parotid gland origin initially presented with SIADH. In the present case, hyponatremia was not persistent even before surgical resection of the pituitary tumor. Kanda *et al.* suggested that the dislocation of the pituitary gland resulted in SIADH based on the preoperative and postoperative MR images [22]. Although it was unclear how the pituitary tumor compressing the pituitary stalk produced SIADH, they speculated either mechanical or chemical stimulation affected the hypothalamo-neurohypophyseal system [22]. Another previous literature reported a case with SIADH caused by a pituitary adenoma with apoplexy [25]. We suspected that severe

pituitary gland compression associated with intratumoral hemorrhage resulted in SIADH in the present case. Hyponatremia was thought to subside after treatment of water intake restriction and induced Na replacement without recurrence, accompanied with gradually resolved pituitary gland compression by hematoma even before adenomectomy.

In conclusion, tumors such as the present case are easily confused with pituitary adenoma or craniopharyngioma. Although rare, metastasis from tumors including those of salivary gland origin should be considered in the differential diagnosis of unusual pituitary tumors.

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