# Refractory Facial Paralysis: A Case Report

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**Abstract-**

***Purpose:*** To present a case of salivary gland malignancy initially mimicking Bell's palsy.

***Case report:*** A 75-year-old woman with hypertension visited our neurological outpatient department, complaining of persistent right facial paralysis for more than a year after oral glucocorticoid therapy with recent development of vertigo and unsteady gait. She was previously diagnosed as having Bell's palsy and was prescribed oral glucocorticoid. However, her right facial muscles were still completely paralyzed, with no signs of improvement. The patient visited the outpatient department of neurology for 3 weeks, seeking treatment for the recent onset of vertigo and ataxia. Brain contrast magnetic resonance imaging (MRI) revealed the right mastoid air cells to be filled with high T2 signal intensity and low T1 signal, with destruction of the bony structure of mastoid, extending to the right jugular bulb. Results obtained from excisional biopsy and pathological analyses were used to diagnose the patient with adenoid cystic carcinoma of the salivary gland. The patient then received a thorough cancer workup and chemoradiotherapy, with the malignancy being under control. However, after a 1-year follow-up, the patient still had permanent right facial palsy.

***Conclusion:*** Salivary gland malignancy should be considered in patients with acute and subacute facial nerve paralysis, in addition to Bell's palsy. Brain imaging with contrast agents should be performed for differential diagnosis.

**Key Words:** Facial palsy; Bell's palsy; Salivary gland malignancy; Adenoid cystic carcinoma.

*Acta Neurol Taiwan 2017;26:68-71*

## INTRODUCTION

Facial palsy is a common condition in the neurological outpatient department and the emergency department. Considering the different courses of symptom progression and the accompanying or following neurological deficits, they have been classified into different etiologies such as aneurysm, infection, malignancy, neuromuscular disease, or other entities [(1)].

The diagnosis of Bell's palsy can be definitive in patients who have a thorough medical history and have undergone physical examinations. However, misdiagnosis can occur in patients who have received classical treatment but with no improvement or complete recovery of facial

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Received March 30, 2017. Revised May 31, 2017.
Accepted July 17, 2017.

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function. All these patients should receive a thorough reevaluation including blood tests and cranial magnetic resonance imaging (MRI) or computed tomographic (CT) scans. We present a case of treatment failure in a patient who was initially diagnosed as having Bell's palsy, but eventually found to have salivary gland malignancy.

***Case***

A 75-year-old woman with hypertension visited our neurological outpatient department complaining of persistent right facial paralysis for more than a year after oral glucocorticoid therapy with recent development of vertigo and unsteady gait. She had had an episode of subacute onset, progressive, diffuse right facial weakness a year ago. Her right facial muscles became completely paralyzed within a week of the onset of symptoms and no other neurological deficit was found initially. The patient was then administered oral glucocorticoid at a dose of 60 mg/day (1 mg·kg-1·day-1) for a total of 7 days, as this was suspected to be a case of Bell's palsy. Facial nerve stimulation and blink reflex studies were performed; these showed significantly reduced amplitudes of compound muscle action potential (CMAP) for the right facial nerve and absence of blink reflex, indicating right facial neuropathy. However, her right facial muscles remained completely paralyzed with no signs of improvement. She visited the outpatient department of neurology for 3 weeks, seeking treatment for recent onset of vertigo and ataxia. There was no taste dysfunction, hearing loss, tinnitus, or oculomotor impairment. For reevaluation, we performed a

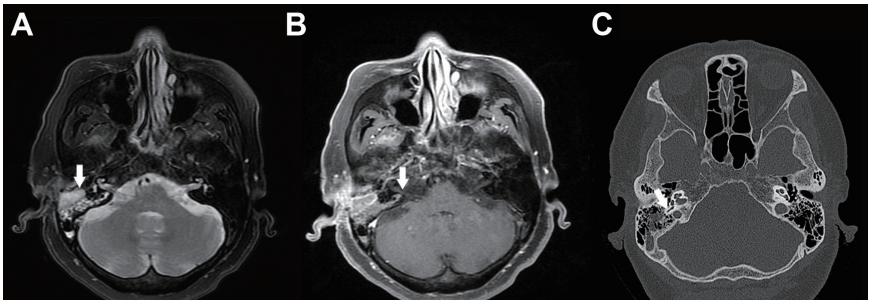


**Figure 1.** Contrast-enhanced brain magnetic resonance imaging (MRI) revealing a T2 hyperintense, heterogeneous contrast enhancing lesion (arrow) involving the right mastoid with destruction of bony structure and extending medially to the jugular bulb (1A) and no direct invasion or compression of the right cochleovestibular nerve (arrow) (1B). Temporal bone computed tomography (CT) showed erosion of the wall of the right posterior semicircular canal (arrow) (1C)

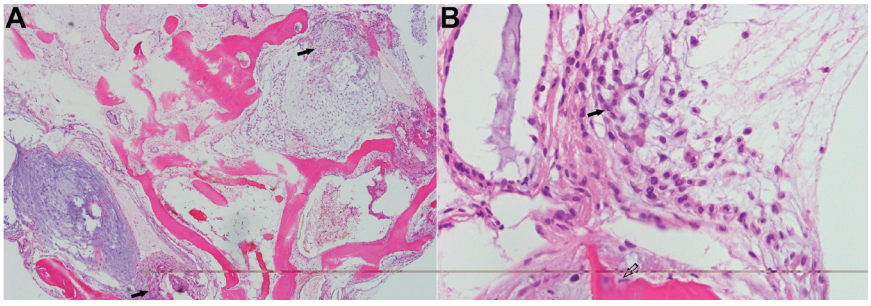


**Figure 2.** The histological section of the tumor showing glandular epithelial cells with myxoid change (black arrow) (hematoxylin-eosin, original magnification ×40) (A) and spindle epithelial cells with mild atypia arranged in the cribriform pattern (black arrow) and bony erosion (white arrow) (hematoxylin-eosin, original magnification ×400) (B).

high contrast brain MRI, which showed the right mastoid air cells to be filled with material having high T2 and low T1 signal intensity, with destruction of the bony structure of mastoid, extending to the right jugular bulb (Figure 1A). There was no direct invasion or compression of the right cochleovestibular nerve (Figure 1B). Temporal bone CT showed erosion of the wall of the right posterior semicircular canal (Figure 1C), which might be the cause of the recent onset of vertigo and ataxia. The diagnosis of adenoid cystic carcinoma of the salivary gland was made based on the results of mastoidectomy with excision of the mastoid tumor and pathological analyses (Figure 2). The patient then received a thorough cancer survey and chemoradiotherapy with the malignancy being under control. However, after a 1-year follow-up, she still had permanent right facial palsy.

## DISCUSSION

Bell's palsy is a common clinical entity, for which the annual incidence rate is between 11.5 and 40.2 cases per 100,000 population [(2)]. Patients with Bell's palsy are often affected with sudden onset, isolated, unilateral, peripheral facial neuropathy, although additional cranial nerve involvement and bilateral facial palsy have also been reported [(3)]. The diagnosis of Bell's palsy is made based on the classic clinical feature of peripheral facial palsy of unknown cause, for which the onset of symptoms is acute and the course is progressive, reaching maximal clinical paralysis within three weeks or less. Early treatment with oral glucocorticoids is recommended. The recovery begins within 3 weeks in approximately 85% of the patients with Bell's palsy, and almost all show considerable recovery in 6 months [(2)]. However, there are numerous etiologies that have a clinical feature resembling Bell's palsy and which may initially involve isolated facial neuropathy. A poor response to treatment or a delayed recovery may prompt a different etiology in addition to Bell's palsy. Some of them could be potentially life-threatening.

It has been reported that benign or malignant neoplasm accounts for about 5% of all cases of peripheral facial palsy; although rarely, some of them may be associated with subacute to acute or even sudden onset facial paralysis [(4)]. However, there are few well-documented cases reported in the literature. Salivary gland tumor is a rare entity accounting for 6-8% of head and neck tumors. The typical clinical presentation of salivary gland neoplasm includes a painless mass or swelling of the involved salivary gland with or without the involvement of cranial nerves and skin. More importantly, a tumor with facial nerve involvement and regional lymph node metastasis is indicative of a malignant neoplasm. Malignant neoplasm of salivary gland is relatively rare compared to other benign conditions. The pathological features of salivary gland malignancy are multifarious, and mucoepidermoid carcinoma and adenoid cystic carcinoma are the most common histologic types, accounting for 33 and 24% of cases respectively [(5)].

In the present case, adenoid cystic carcinoma of the salivary gland was associated with a subacute onset, isolated right facial palsy, in which the facial muscles became completely paralyzed within a week without a palpable mass, salivary gland swelling, or skin involvement. The patient was initially diagnosed as having Bell's palsy based on the clinical features and treated with oral glucocorticoid. However, there was no improvement of the facial weakness. A contrast-enhanced brain MRI scan was performed 1 year later and the diagnosis of salivary gland tumor was made after considerable delay. This case illustrates the challenge of early diagnosis of salivary gland malignancy without a cranial image study, in patients who have no palpable mass lesion. The only symptom in this case was unilateral facial nerve invasion that mimicked Bell's palsy.

In conclusion, although malignant neoplasm of the head and neck is not a common clinical entity in patients with acute facial palsy, a thorough medical history and physical examination could help in differential diagnosis. However, some patients could have an occult malignancy with no other symptoms or signs other than facial nerve paralysis. In these patients, a poor response to steroid treatment may be observed, with a delayed recovery of facial function over 6 months. In such cases, reevaluation should be performed, including a head and neck MRI with enhanced contrast.

## ACKNOWLEDGMENT

None.

## REFERENCE


1. Hohman MH, Hadlock TA. Etiology, diagnosis, and management of facial palsy: 2000 patients at a facial nerve center. The Laryngoscope. 2014;124 (7):E283-E93.
2. Eviston TJ, Croxson GR, Kennedy PG, Hadlock T, Krishnan AV. Bell's palsy: aetiology, clinical features and multidisciplinary care. J Neurol Neurosurg Psychiatry. 2015:jnnp-2014-309563.
3. Benatar M, Edlow J. The spectrum of cranial neuropathy in patients with Bell's palsy. Archives of internal medicine. 2004;164(21):2383-5.
4. Quesnel AM, Lindsay RW, Hadlock TA. When the bell tolls on Bell's palsy: finding occult malignancy in acute-onset facial paralysis. American journal of otolaryngology. 2010;31(5):339-42.
5. Jones A, Craig G, Speight P, Franklin C. The range and demographics of salivary gland tumours diagnosed in a UK population. Oral oncology. 2008;44(4):407-17.