

case report

Pineal gland metastasis of auricular squamous cell carcinoma: an unusual case and literature review

Ozgun Oztekin¹, Recep Savas², Ebru Ozan¹, Melda Apaydin³,
Öyküm Yaşar¹, Zehra Hilal Adibelli¹

¹Izmir Education and Research Hospital, Radiology Department,

²Ege University Faculty of Medicine, Radiology Department,

³Ataturk Education and Research Hospital, Radiology Department, Izmir, Turkey

Background. The pineal gland is an unusual site for metastasis, and most metastatic pineal lesions are asymptomatic. Metastases to the pineal gland from skin cancer are extremely rare and reported mostly on autopsy series. Squamous cell carcinoma is the second most common type of skin cancer that occurs on the external ear. Auricular squamous cell carcinoma is an invasive and destructive tumour, and may cause hearing problems by local extension to the auditory canal. The vast majority of squamous cell carcinomas of the auricular region metastasize to the lung, bone, and brain.

Case report. We report the case of a patient with a giant squamous cell carcinoma of the auricula with extension deep into the temporal bone, metastasizing to the lung and pineal gland.

Conclusions. A metastasis should be considered as a possible cause, when encountering a mass in the pineal region, especially in elderly patients with a known primary cancer.

Key words: pineal gland; metastasis; squamous cell carcinoma

Introduction

Metastases to the pineal gland region are rare, accounting for only 0.3% of all brain metastasis in a series from Japan¹ but ranges from 1.8% to 4% in other series. The most frequently reported sites of primary tumour involvement included breast and

bronchogenic carcinomas, with melanotic, renal cell, pancreatic, ovarian, gastric, and frontal sinus malignancies being less common. To date there have been fewer than 100 reports of metastatic lesions in the pineal region.¹⁻⁶ In a large survey from Japan, metastases to the pineal region were found in 23 patients and the primary tumour sites were melanoma in five cases found at autopsy.¹ Cutaneous squamous cell carcinoma (SCC) is a common skin cancer. The most common sites of metastases are regional lymph nodes, lung, liver, brain, skin and bone. However, until now metastases to the pineal gland were not reported. Here

Received 26 January 2009

Accepted 4 May 2009

Correspondence to: Dr. Ozgur Oztekin, Albayrak mavişehir evleri, Yalı mahallesi, 6525sok. No:35 daire no:31 Karşıyaka, Izmir, Turkey. Phone: +9053 2333 0201, +9050 5376 7387; Fax: +9023 2250 5050; E-mail: oztekinogur@gmail.com

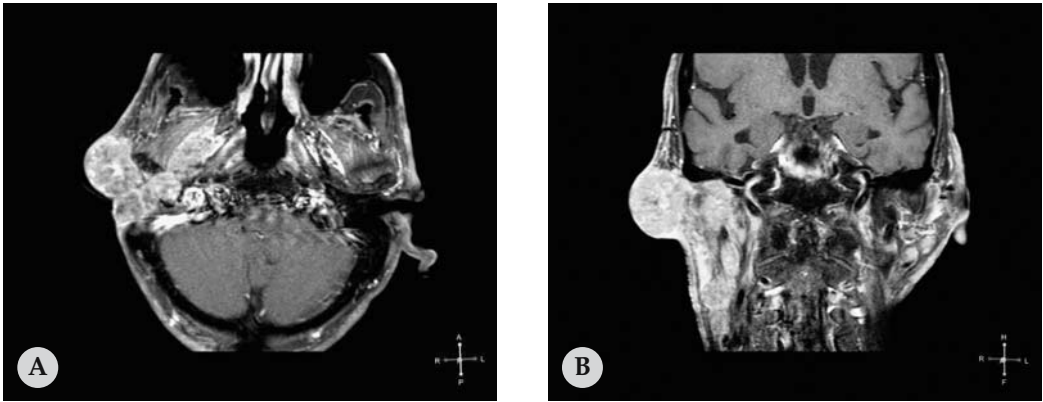


Figure 1. Cerebral MRI scans. (A) Axial spectral presaturation with inversion recovery (SPIR) image clearly demonstrates tumour invasion from the external auricular part deep into the temporal bone. (B) Coronal SPIR image delineate local aggressive extension of the squamous cell carcinoma to the cervical lymph nodes. Diffuse homogeneous contrast enhancement is seen after intravenous contrast injection in SPIR series.

we present, to the best of our knowledge, the first case of a skin squamous cell carcinoma that metastasized to the pineal gland.

Case report

A 75-year-old male farmer with progressive hearing loss and chewing problems was evaluated at the ear, nose, and throat outpatient clinic. The patient's history included the diagnosis of squamous cell carcinoma of the right auricula 15 years ago; the lesion was excised several times but recurred. Metastases to the right submandibular gland and right upper deep cervical chain lymph nodes were excised 6 years ago. Ten months previous, the patient was admitted to the hospital with haemoptysis and nonproductive cough, and multiple bilateral metastatic lung lesions were diagnosed on computerized tomography (CT) of the thorax. At that time, the patient underwent abdominal ultrasonography and cranial magnetic resonance imaging (MRI) to evaluate the possibility of metastases to other sites, and no other abnormality was found.

The patient had no other neurological signs or symptoms at the time of admis-

sion. On physical examination, he had an erythematous skin lesion on his right auricula and lesions of the lower lip and right hand. The auricular lesion had eroded the ear and tragus and obstructed the external acoustic meatus. Cranial MRI was repeated, and its results showed that the auricular lesion invaded the temporal bone from the mastoid portion deep into the petrosal apex. Mastoid aeration was lost, and all parts of the temporal bone had an expansible character. Expansion in the petrosal part of the temporal bone exerted pressure on the muscles of mastication (Figure 1). In addition, an abnormal contrast-enhancing mass was observed in the pineal gland that was not seen on the previous cranial MRI (Figure 2 a,d). The well-defined mass was isointense on T1-weighted imaging and uniformly hypointense on T2-weighted imaging (Figure 2 b,c).

Discussion

Pineal metastases are generally considered unusual and incidental events that occur late in the course of widely metastatic systemic cancer. The first report of pineal

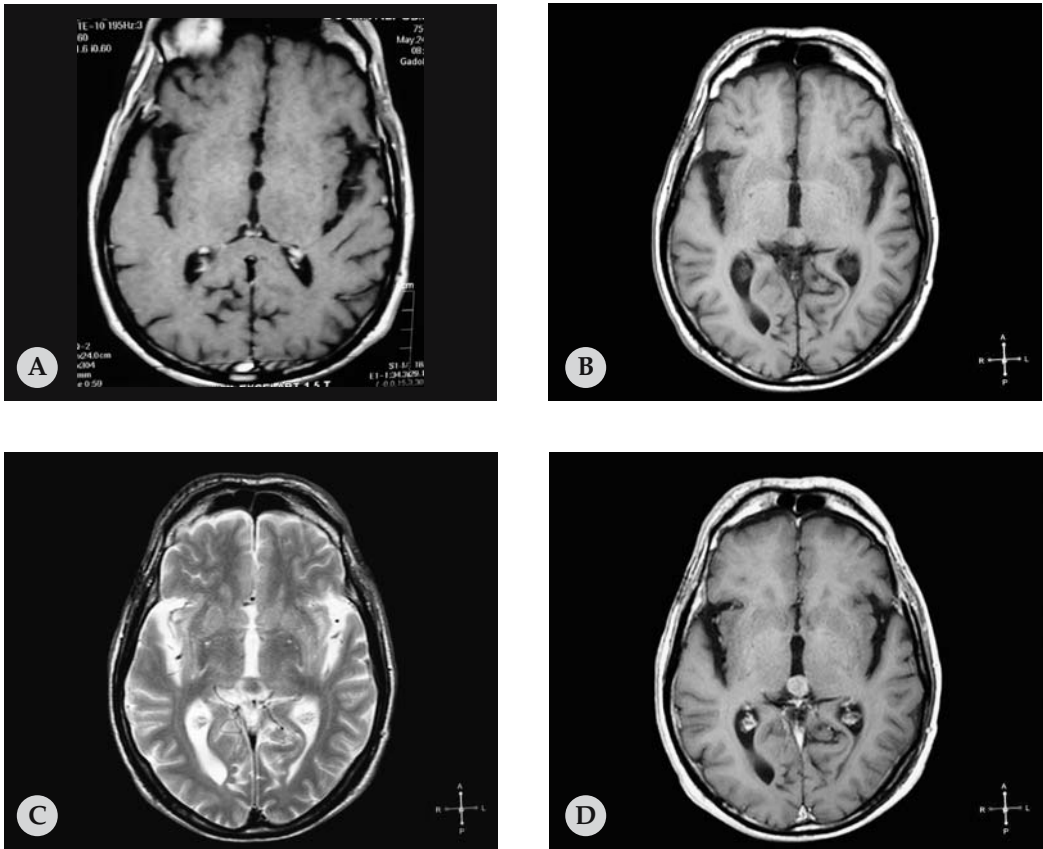


Figure 2. (A) Axial postcontrast T1-weighted image was normal when the patient was admitted to the hospital 10 months earlier. (B) Pineal mass is isointense on T1-weighted, (C) and uniformly hypointense on T2-weighted images. (D) Postcontrast T1-weighted axial images demonstrate uniform contrast enhancement.

gland metastasis was described in 1858 by Forster.¹ The most commonly reported primary tumours are carcinomas of the lung, breast, stomach, oesophagus, rectum, and kidney.² There also have been case reports of pineal region metastases from haematological malignancies and melanomas.² In a large number of older cases, metastases to the pineal gland likely metastases to the pituitary one were found at autopsy³⁻⁷, but more recently, imaging, especially MRI, has markedly improved the earlier detection of such lesions.^{2,8-12} Ortega *et al.*⁴ found a 3.8% prevalence (5 cases) of pineal gland metastases in a cohort of 130 patients with the widespread neoplastic disease. Three of

these cases had isolated pineal lesions, all of which had grossly replaced pineal tissue. The remaining 2 cases revealed other sites of intracranial involvement with only a minimal pineal gland replacement. Andrew *et al.*¹² observed a 5% prevalence in a series of patients referred for the surgical management of pineal gland tumours.

In a large survey of 7807 intracranial metastatic tumours in Japan,¹ metastases to the pineal region were found in 48 patients, for a frequency of 0.3%. Cases constituting a solitary mass, detectable by CT or MRI, were even less frequent. The primary tumour sites were lung (22 cases), breast (9 cases), malignancies of the gastrointestinal

tract (stomach, 3; oesophagus, 2; colorectal, 1), skin (melanoma, 5), hematologic system (multiple myeloma, 2; plasma cell leukaemia, 1; lymphoma, 1), kidney (clear cell carcinoma, which can also metastasize to the pineal gland),⁷ and frontal sinus.¹

Squamous cell carcinoma is the second most common skin cancer, and it represents 20% of all cutaneous malignancies.¹³ Certain cervicofacial regions, such as the ear and lower lip, are more prone to developing squamous cell carcinoma.¹⁴ Squamous cell carcinoma of the external ear is a common diagnosis, and it occurs with greatest frequency on the superior or posterior portions of the helical rim. Several studies have demonstrated a higher recurrence rate as well as a poorer prognosis for squamous cell carcinoma of the external ear, compared to other cutaneous squamous cell carcinoma locations. Metastases from squamous cell carcinoma of the external ear occur in 5% to 18% of cases, compared to metastatic disease in only 0.5% to 2% of cutaneous squamous cell carcinoma occurring elsewhere.¹⁵

The lymphatic drainage of the auricula is plentiful, with at least 3 distinct lymphatic territories draining the skin of the auricula. It is useful to consider auricular squamous cell carcinoma separately from other high-risk squamous cell carcinoma when looking for potential markers of metastatic potential.¹⁵ The parotid gland and the upper deep cervical chain are the most common sites of metastases.¹⁵ The postauricular nodes are also frequently involved.¹⁵ Auricular squamous cell carcinoma is associated with a significant mortality at 6.2%, which is usually due to the failure of control at the level of the local lymph node basins. Freedlander *et al.*¹⁶ reported that tumours with a diameter over 3 cm have a metastatic potential of over 44%.

In the present patient, beside local recurrences of squamous cell carcinoma, submandibular gland and upper deep cervical

chain lymph node metastases were found during the course of the disease, all of which caused local effects in the head and neck region. This patient also had widespread lung metastases that caused systemic complaints. Interestingly, the patient did not exhibit any neurological symptoms related to pineal metastases at the time of admission.

History of malignancy is the most important factor in differentiating metastatic intracranial disease from a primary lesion. In patients with a known history of malignant neoplasm, approximately 90% of supratentorial lesions represent metastases.¹¹ Young adults and children usually are those affected by primary pineal tumours. Our patient had a primary squamous cell carcinoma of auricular skin and was elderly, both of which are consistent with metastases to the pineal gland. The results of the patient's previous cranial MRI were normal when his lung lesions were diagnosed 10 months earlier. All of these factors led us to a diagnosis of metastatic disease, rather than a primary pineal tumour.

In conclusion, when encountering a mass in the pineal region, a metastasis should be considered as a possible cause, especially in elderly patients and those with a history of malignancy. Although such metastases are rare, the present patient provides information that might be useful for diagnosis.

References

1. Report of brain tumor registry of Japan (1969-1993). *Neurol Med Chir (Tokyo)* 2000; **40 Suppl**: 24.
2. Kakita A, Kobayashi K, Aoki N, Eguchi I, Morita T, Takahashi H. Lung carcinoma metastasis presenting as a pineal region tumor. *Neuropathology* 2003; **23**: 57-60.
3. Chason JL, Walker FB, Landers JW. Metastatic carcinoma in the central nervous system and dorsal root ganglia. A prospective autopsy study. *Cancer* 1963; **16**: 781-7.

4. Ortega P, Malamud N, Shimkin MB. Metastasis of carcinoma to the pineal body. *Arch Pathol* 1951; **52**: 518-28.
5. Halpert B, Erickson EE, Fields WS. Intracranial involvement from carcinoma of the lung. *Arch Pathol* 1960; **69**: 93-103.
6. Vaquero J, Martínez R, Magallón R, Ramiro J. Intracranial metastases to the pineal region. Report of three cases. *J Neurosurg Sci* 1991; **35**: 55-7.
7. Lauro S, Trasatti L, Capalbo C, Mingazzini PL, Vecchione A, Bosman C. Unique pineal gland metastasis of clear cell renal carcinoma: case report and review of the literature. *Anticancer Res* 2002; **22**: 3077-9.
8. Bišof V, Juretić A, Šarić N, Melada A, Perković Z, Radoš M, et al. Pituitary metastasis of renal cell carcinoma: a case report. *Radiol Oncol* 2008; **42**: 225-31.
9. Kanai H, Yamada K, Aihara N, Watanabe K. Pineal region metastasis appearing as hypointensity on T2-weighted magnetic resonance imaging-case report. *Neurol Med Chir (Tokyo)* 2000; **40**: 283-6.
10. Brasseur P, Sukkarieh F, Dupont H, Brohéé D. Pineal body metastasis. *J Belge Radiol* 1994; **77**: 162-3.
11. Weber P, Shepard KV, Vijayakumar S. Metastases to pineal gland. *Cancer* 1989; **63**: 164-5.
12. Andrew B, Lassman AB, Bruce JN, Fetell MR. Metastases to the pineal gland. *Neurology* 2006; **67**: 1303-4.
13. Wade TR, Ackerman AB. The many faces of basal cell carcinoma. *J Dermatol Surg Oncol* 1978; **4**: 23-8.
14. Siegle RJ, MacMillan J, Pollack SV. Infiltrative basal cell carcinoma: a nonsclerosing subtype. *J Dermatol Surg Oncol* 1986; **12**: 830-6.
15. Clark RR, Soutar DS. Lymph node metastases from auricular squamous cell carcinoma. A systematic review and meta-analysis. *J Plast Reconstr Aesthet Surg* 2008; **61**: 1140-7.
16. Freedlander E, Chung FF. Squamous cell carcinoma of the pinna. *Br J Plast Surg* 1983; **36**: 171-5.