Lung adenocarcinoma with initial symptom of occipital condyle syndrome – a case report

Adenokarcinom plic se syndromem okcipitálního kondylu jako počátečním příznakem – kazuistika

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Summary

Background: A metastasis to the basilar bones, including the occipital bone, is extremely rare. Such metastases can result in hypoglossal nerve dysfunction, which poses diagnostic challenges. We report a case of lung cancer presenting with hypoglossal nerve dysfunction caused by metastasis to the posterior condyle of the occipital bone. ***Case:*** A 52-year-old man was referred to our hospital due to hypoglossal nerve dysfunction and occipital pain. Imaging studies revealed a metastatic lesion in the posterior condyle of the occipital bone and a 12-mm nodule in the apex of the left lung. Histopathological analysis confirmed stage IVB lung adenocarcinoma without actionable driver mutations or significant PD-L1 expression. The patient underwent radiation therapy targeting the occipital lesion, followed by systemic therapy with carboplatin, paclitaxel, bevacizumab, and atezolizumab. Maintenance therapy with bevacizumab and atezolizumab was continued. After 20 months of follow-up, the patient remained disease-free, with resolution of hypoglossal nerve dysfunction. Imaging confirmed ossification at the site of the bone metastasis, indicating a favorable response to treatment. ***Conclusion:*** Although such cases are extremely rare, this report provides valuable insight into the management of patients with similar metastases. Early diagnosis and a multidisciplinary approach, including chemotherapy, immunotherapy, and radiation therapy, are essential for improving outcomes in patients with rare metastases involving the posterior condyle of the occipital bone.

Key words

occipital condyle syndrome – occipital bone – hypoglossal nerve diseases – adenocarcinoma of lung – skull base neoplasms – bone metastasis – immune checkpoint inhibitors – radiotherapy

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Souhrn

Východiska: Metastázy do kostí báze lební, vč. týlní kosti, jsou velmi vzácné. Tyto metastázy mohou vést k dysfunkci hypoglosálního nervu, což představuje diagnostický problém. Uvádíme případ karcinomu plic s dysfunkcí hypoglosálního nervu způsobenou metastázami do zadního kondylu týlní kosti. ***Případ:*** Dvaapadesátiletý muž byl odeslán do naší nemocnice kvůli dysfunkci hypoglosálního nervu a bolestem v týle. Zobrazovací techniky odhalily metastatickou lézi v zadním kondylu týlní kosti a 12mm uzlík v levém plicním hrotu. Histopatologické vyšetření potvrdilo adenokarcinom plic stadia IVB bez řídicích mutací nebo významné exprese PD-L1. Pacient podstoupil radioterapii zaměřenou na okcipitální lézi, po níž následovala systémová léčba karboplatinou, paklitaxelem, bevacizumabem a atezolizumabem. Udržovací léčba pokračovala podáváním bevacizumabu a atezolizumabu. Po 20 měsících sledování zůstal pacient bez onemocnění, přičemž dysfunkce hypoglosálního nervu vymizela. Zobrazovací techniky potvrdily osifikaci v místě kostní metastázy, což svědčí o příznivé odpovědi na léčbu. ***Závěr:*** Ačkoli jsou takové případy extrémně vzácné, tento článek poskytuje cenné informace o léčbě pacientů s podobnými metastázami. Včasná diagnóza a multidisciplinární přístup, zahrnující chemoterapii, imunoterapii a radioterapii, jsou nezbytné pro zlepšení výsledků u pacientů se vzácnými metastázami postihujícími zadní kondyl týlní kosti.

Klíčová slova

syndrom okcipitálního kondylu – týlní kost – onemocnění hypoglosálního nervu – adenokarcinom plic – novotvary báze lební – kostní metastázy – inhibitory imunitních kontrolních bodů – radioterapie

Introduction

Some patients with lung cancer develop symptoms caused by distant metastatic lesions [1]. Furthermore, when the primary lesion is small and located in a site challenging for tissue sampling, diagnosis can be difficult, potentially delaying the initiation of appropriate treatment. Here, we describe a patient with lung cancer who initially presented with hypoglossal nerve dysfunction due to metastasis to the posterior condyle of the occipital bone. The primary lesion was a very small adenocarcinoma located in the lung apex. Considering the size and location of the primary tumor, a surgical approach was required to obtain pathologic tissue for histological diagnosis. Treatment included radiation therapy to the posterior condyle of the occipital bone, chemotherapy, and immune checkpoint inhibitor, which proved effective. In this patient, accurate diagnosis and appropriate treatment contributed to long-term survival. The clinical course of this patient offers valuable insights for managing future patients with similar presentations.

Case report

A 52-year-old man experienced abnormal tongue movements and occasional pain radiating from the left occipital region to the neck. These symptoms continued for two months, prompting him to visit the neurosurgery department of a hospital. A head CT scan showed a 13-mm ring-shaped nodular lesion with enhancement in the right parietal lobe (Fig. 1) and osteolytic changes in the left occipital condyle (Fig. 2A). The lesions were suspected to be metastatic, so the patient was referred to our hospital for further evaluation. A plain chest radiograph and chest CT scan showed a 12-mm nodule in the apex of the left lung (Fig. 3). On physical examination at the time of admission to our hospital, no superficial lymphadenopathy was detected, and cardiac and respiratory sounds were normal. However, neurological examination revealed that the tongue could not be protruded. Fasciculations were observed on the tongue and the left side was atrophic and deviated to the left. Serum carcinoembryonic antigen level was elevated to 19.3 ng/mL. Fluorodeoxyglucose positron emission tomography (FDG-PET) imaging revealed FDG uptake in the left apical nodule and the left occipital condyle (Fig. 2B, 3C). Due to the difficult obtaining of tissue samples for histological diagnosis via bronchoscopy or percutaneous biopsy, surgical sampling was performed. Histological analysis of the biopsy specimen revealed trabecular, alveolar, and papillary proliferation, as well as alcian blue-positive cells, consistent with adenocarcinoma. Immunohistochemical staining was positive for pancytokeratin, CK-7, and TTF-1 confirming a diagnosis of lung adenocarcinoma, T2aN0M1b, stage IVB. No driver gene mutations were identified, and PD-L1 expression was < 1%. The patient underwent radiation therapy for the metastatic brain lesions and bone metastases in the occipital condyle, followed by four courses of carboplatin, paclitaxel, bevacizumab, and atezolizumab. Maintenance therapy with bevacizumab and atezolizumab continued for 12 months. A CT scan taken 14 months after treatment initiation showed significant shrinking of the primary lesion and brain metastasis, along with ossification and reduction of osteolytic lesion in the occipital condyle (Fig. 4). Tongue movement and pain in the left occipital region and neck improved, and no residual disability was observed. Twenty months after the treatment began, there are no signs of recurrence, and maintenance therapy with bevacizumab and atezolizumab is ongoing.

Discussion

Hypoglossal nerve palsy is a paralysis which primarily affects the tongue muscles. It is characterized by hemi-atrophy of the tongue, fasciculations, deviation of the tongue to the affected side during anterior protrusion, and dysarthria. The hypoglossal nerve passes through the hypoglossal canal and the anterior condylar foramen to reach the outside of the skull. Metastasis to the occipital condyle can disrupt this pathway, leading to ipsilateral hypoglossal nerve palsy [2]. In most cases, hypoglossal nerve palsy is unilateral and often occur in combination with glossopharyngeal, vagal, and accessory nerve palsies [3]. In a retrospective study by Stino et al. involving 245 patients with hypoglossal nerve palsy, metastasis from malignant tumors

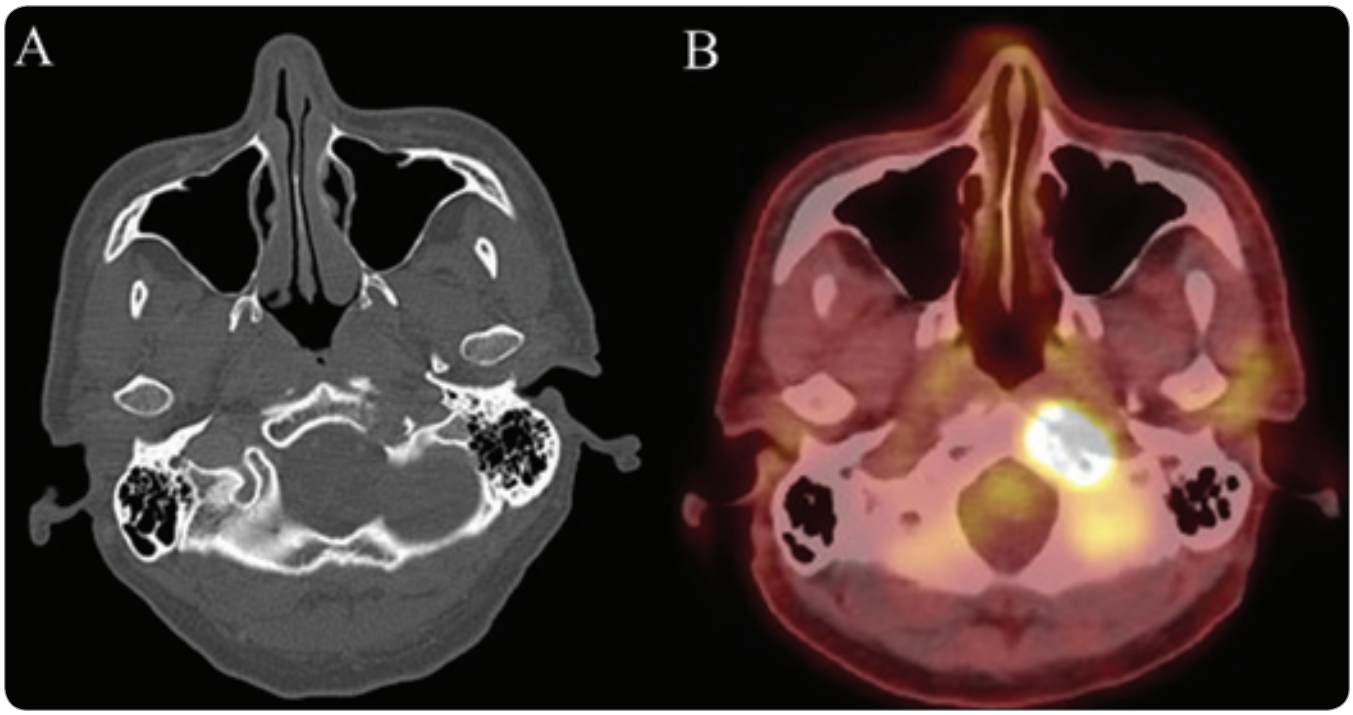

Fig. 2. (A) Head CT scan taken at the time of the onset showed an osteolytic change in the left occipital condyle; (B) PET/CT revealed uptake of fluorodeoxyglucose at the osteolytic change in the left occipital condyle.

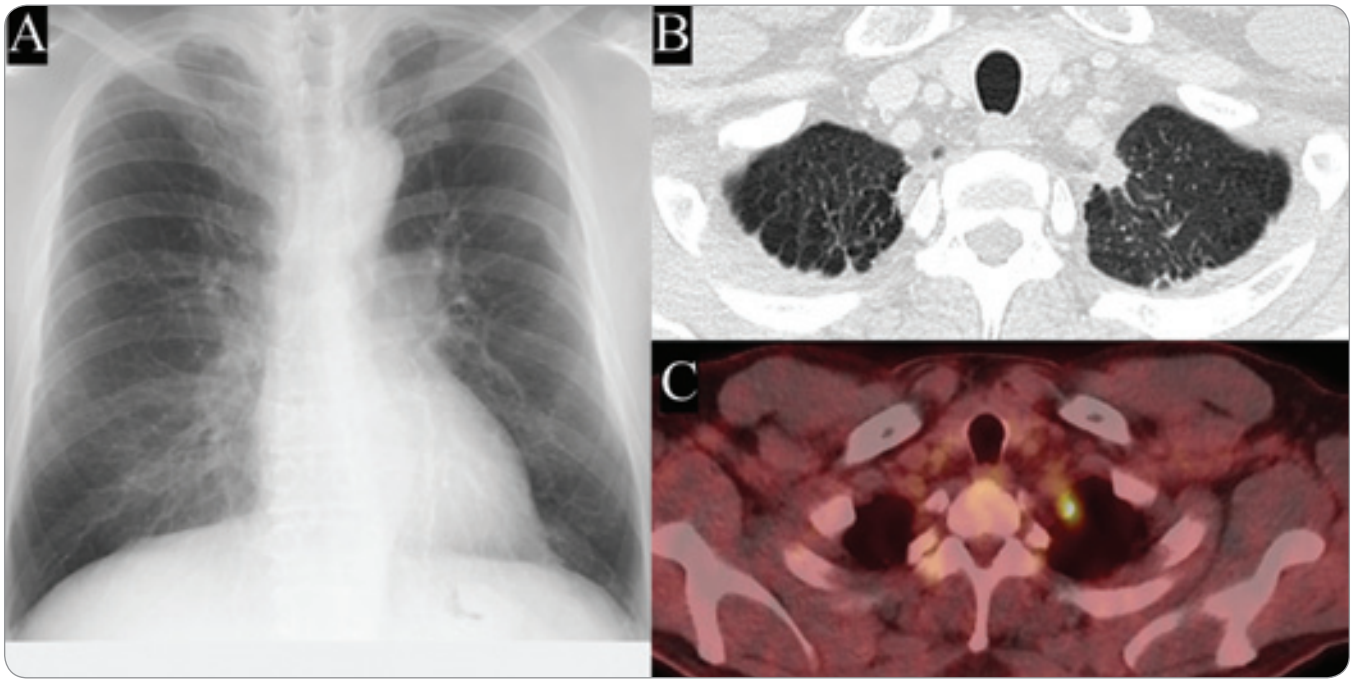

Fig. 3. (A) Plain chest radiograph; (B) chest CT scan. Both images showed a 12-mm nodule in the apex of the left lung. (C) PET/CT revealed uptake of fluorodeoxyglucose at the nodule in the apex of the left lung.

was identified as the fourth most common etiology, accounting for 13% of the examined cases [4]. One cause of hypoglossal neuropathy is occipital condyle syndrome (OCS), a condition characterized by headache and hypoglossal neuropathy due to lesions in the occipital condyle [5,6]. OCS can be caused by metastatic lesions in the occipital condyle.

Tumors that cause OSC include prostate cancer [7] and breast cancer [8]; however, to the best of our knowledge, only a few cases of OSC associated with lung cancer have been reported in the

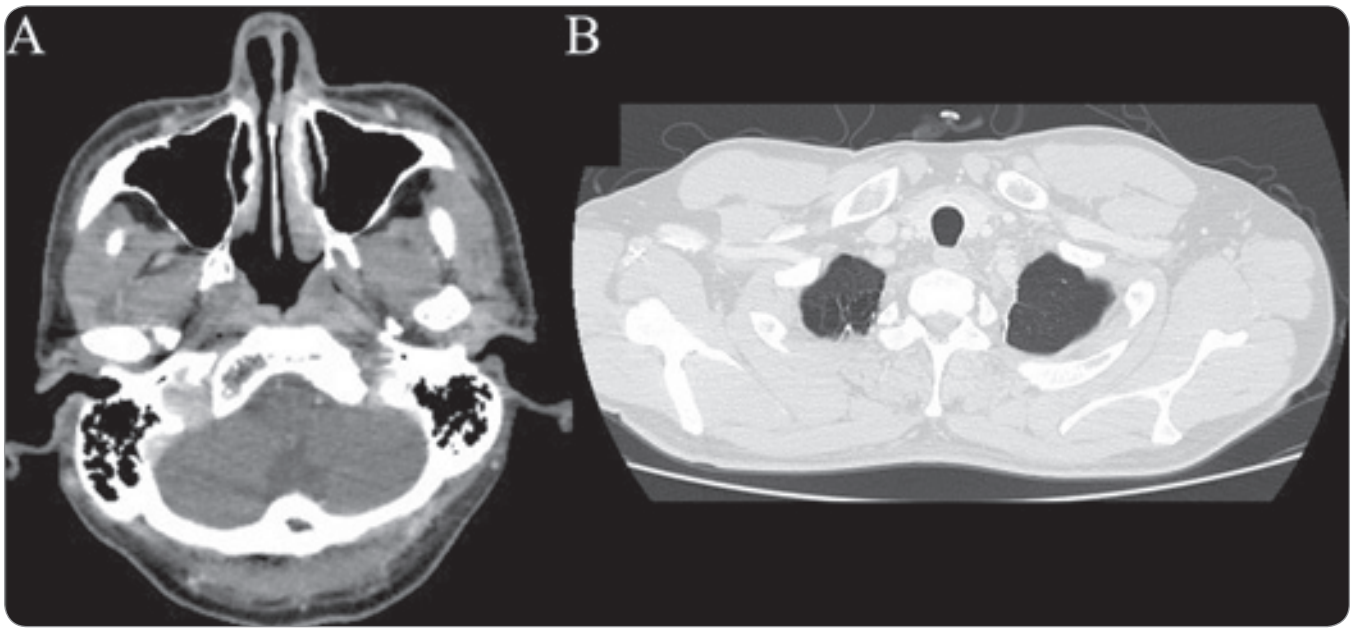

Fig. 4. (A) CT scan taken 14 months after the start of the treatment revealed disappearance of the primary lung lesion; (B) the osteolytic lesion in the occipital condyle had shrunk and showed ossification.

literature [2,9,10]. In the presented patient, the tongue could not move as desired, and there was pain radiating from the left occipital region to the neck, which is typical of OCS. The most critical diagnostic takeaways from this patient's clinical course were the importance of taking a detailed medical history, performing a thorough physical examination, and considering OCS as a differential diagnosis. OCS is typically observed during cancer progression rather than as an initial presentation [11] and is associated with a very poor prognosis [11,12]. According to Laigle-Donadey et al., the overall survival from the time of skull metastasis diagnosis was 5 months [12]. Gamma knife treatment is gaining attention as a local control therapy for OCD [13]. Meanwhile, in systemic therapy for advanced lung cancer, immune checkpoint inhibitors have revolutionized treatment outcomes. In this case, our patient received radiation therapy to the metastatic region in the occipital condyle, along with chemotherapy and immune checkpoint inhibitors, and has survived without recurrence for 20 months since the start of his treatment. The hypoglossal nerve impairment symptoms have also improved, and CT scans have confirmed the ossification at the metastatic site in the occipital condyle. Our patient is currently undergoing maintenance therapy with bevacizumab and atezolizumab. To the best of our knowledge, no other cases of metastatic OCS have been reported to achieve such a long survival time. We believe that the clinical course in our patient offers valuable insights into the treatment of future patients with similar presentations of metastatic OCS from lung cancer.

Conclusions

Although extremely rare, lung cancer patients can develop symptoms of OCS. Therefore, careful medical history taking and appropriate evaluation of symptoms are essential to ensure accurate diagnosis and optimal treatment. In addition, treatments including immune checkpoint inhibitors may contribute to long-term survival and should be considered as potential options when selecting treatment strategies.

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