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Case Report

# Olfactory neuroblastoma pulmonary metastasis presenting as a solitary pulmonary mass in an adult: A case report with pathologic correlation

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ABSTRACT

Pulmonary metastasis of an esthesioneuroblastoma is uncommon. In this report, we present a case of an esthesioneuroblastoma pulmonary metastasis in an adult. A 39-year-old man treated with surgical resection of olfactory neuroblastoma was found to have a solitary pulmonary mass on a surveillance computed tomography scan performed 5 years after undergoing primary surgery and radiation treatment. He underwent lobectomy, and histopathology revealed esthesioneuroblastoma metastasis. To our knowledge, no case of esthesioneuroblastoma metastasis presenting as a solitary pulmonary nodule has been described in peer-reviewed literature.



## Introduction

Esthesioneuroblastoma (ENB) also known as olfactory neuroblastoma is an uncommon neoplasm arising from nasal-olfactory epithelium. Although Berger and Luc initially described this entity in 1924, there is very limited literature regarding the incidence and spread of the disease [1,7]. It usually presents as a locally invasive tumor with a high propensity of regional sinonasal involvement. Aggressive treatment strategy with surgery and radiation therapy has resulted in good local control of disease, which has shown to provide the best disease-free survival and overall survival [2–4]. However, regional and distant metastasis following primary treatment is not uncommon, preferred sites including neck, brain, spine, and lungs [5]. Lung metastasis is a very rare occurrence in adults; fewer than 10 reported cases diagnosed by tissue biopsy have been documented [13]. We describe a case of an ENB pulmonary metastasis presenting as a solitary pulmonary nodule in an adult. The patient underwent resection of the pulmonary metastatic mass.

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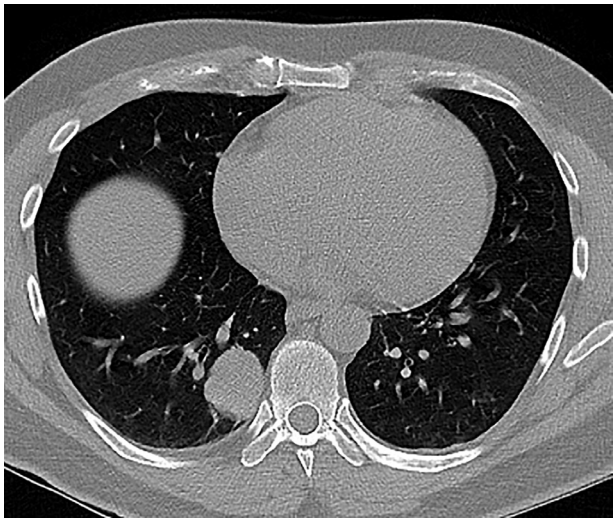

**Fig. 1 – Axial CT chest with contrast shows 4 cm right lower lobe solid pulmonary mass.**

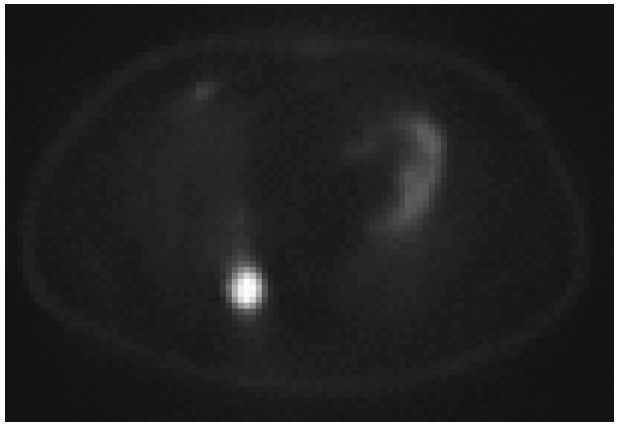

**Fig.2 – PET CT chest shows FDG avid (6 SUV) right lower lobe solid pulmonary mass and enlarged right paratracheal lymph node.**

## Case report

A 59–year-old-man was admitted to the hospital following a minor motor vehicle accident. Past medical history was significant for resection of olfactory neuroblastoma and postoperative proton therapy 5 years back. He had no surveillance imaging performed since initial treatment. Whole-body computed tomography (CT) was performed as part of the trauma protocol. CT chest revealed no acute traumatic injury, but showed a 4 cm well-defined solitary pulmonary mass in the medial basal segment of the right lower lobe (Fig. 1) and enlarged right paratracheal lymph node. Positron Emission Tomography (PET) CT showed Fluorodeoxyglucose (FDG) avid right lower lobe mass and right paratracheal node, both showing 6 Standard Uptake Value (SUV) (Fig. 2). There was no other FGD avid uptake to suggest extrathoracic metastasis or local recurrence. Initial pretreatment contrast-enhanced MRI reveals enhancing tumoral lesion originating from the paranasal sinuses with intracerebral extension through the cribriform plate (Fig. 5).

He subsequently underwent a CT-guided biopsy of the right lower lobe and endobronchial ultrasound-guided biopsy of the right paratracheal lymph node. The pulmonary mass and right paratracheal node revealed a histopathologic diagnosis of ENB metastasis. He underwent right lower lobectomy, and was commenced on chemotherapy (carboplatin and etoposide). Two-month follow-up CT chest showed complete resolution of the right paratracheal lymph node.

## Pathology findings

Microscopic examination of the dominant right lower lobe mass demonstrated a peripherally located neuroen-

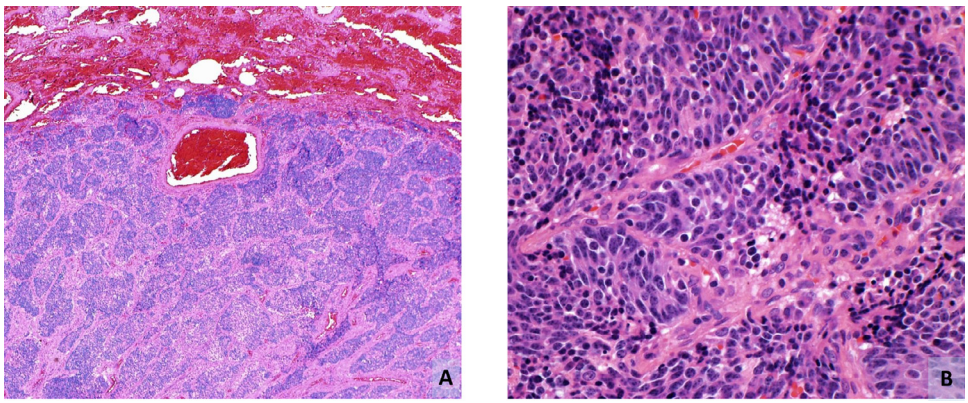


**Fig. 3 – (A) The solitary pulmonary nodule demonstrating nests and trabeculae of tumor cells (H&E, 4×). (B) High-grade areas of the tumor with neoplastic cells demonstrating high nuclear/cytoplasmic ratios, increased mitotic activity, and increased apoptosis (H&E, 40×).**

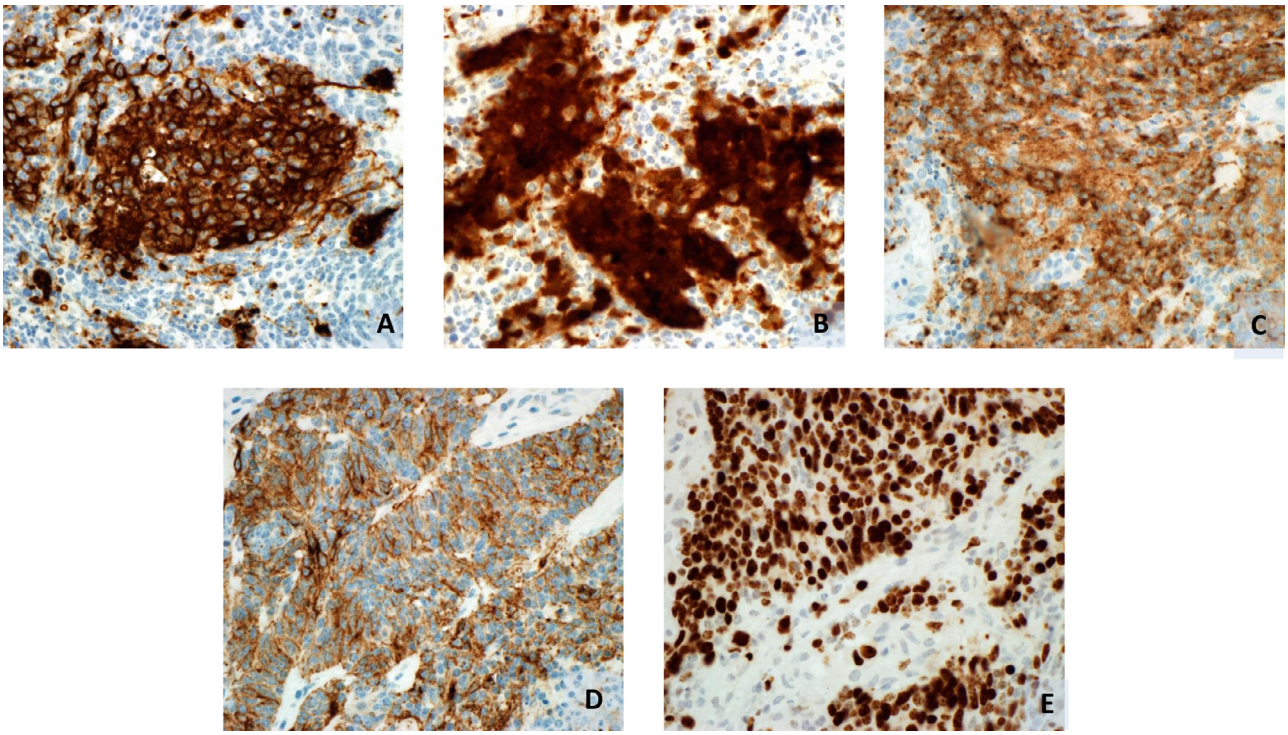


**Fig. 4 – Tumor cells showed diffuse immunohistochemical staining for (A) CD56, (B) synaptophysin, and (C) calretinin (all 40×) with more focal positivity for (D) pan-cytokeratin (40×). Ki-67 immunohistochemical stain (E) demonstrated a markedly elevated labeling index (40×).**

docrine neoplasm that demonstrated areas of both low and high-grade differentiation (Fig. 3). The high-grade areas showed nests and trabeculae of neuroendocrine cells with high nuclear/cytoplasmic ratios, prominent nucleoli, focal coagulative tumor necrosis, and an increased mitotic rate (20 mitoses/10 high power fields). A battery of immunohistochemical tests was performed and demonstrated the tumor was diffusely positive for CD56, synaptophysin, and calretinin (Figs. 4A respectively) with focal positivity for pancyto-keratin and a Ki-67 labeling index of greater than 50% (Figs 4). After review of the pathology of the patient's initial skull base tumor, the final diagnosis was that the lung tumor was a solitary metastasis from the patient's original ENB, rather than new primary such as a pulmonary large cell neuroendocrine tumor.

## Discussion

ENB is a rare malignant tumor that accounts for 6% of malignant nasal cavity neoplasms [6]. The Kadish system is used to stage ENB based upon spread of tumor, with stage A confined to the nasal cavity, stage B involving the nasal sinuses, and stage C tumors spreading into the middle cranial fossa and retrobulbar orbit [7]. Due to a high incidence of local tumor recurrence following surgical treatment, radiation and adjuvant chemotherapy have become an integral treatment strategy for ENB patients [8,9]. Cervical metastasis can present in the early stages or later with an incidence of 10%-30% at the time

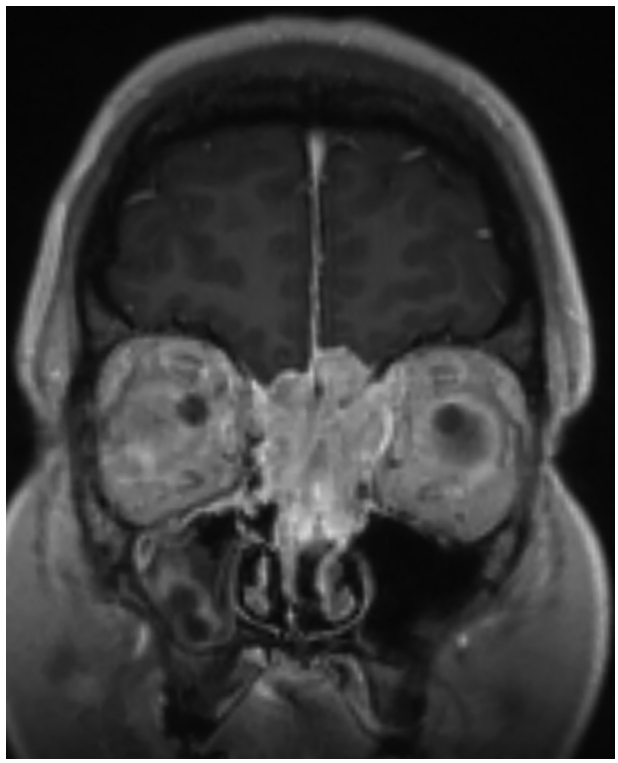

**Fig. 5 – Contrast-enhanced coronal-MRI reveals enhancing tumoral lesion originating from the paranasal sinuses with intracerebral extension through the cribriform plate.**

of diagnosis [10]. Neck recurrence can occur between 2 and 10 years from the initial diagnosis [8,10,14]. The incidence of distant metastasis (such as lung, brain, bone) occurs in 12%-25% of patients [11]. Fewer than 10 cases with pulmonary metastasis have been documented in the literature. Tural et al describe the occurrence of distant metastasis to meninges, spinal cord, bone, and lung between 8.5 and 124 months with a median time of 8.4 months.

Lung metastasis of primary malignant tumors most often occurs by hematogenous spread followed by lymphatic and endobronchial dissemination. Common extrathoracic sites of primary tumors include breast, kidney, colon, thyroid, pancreas, prostate, stomach, and uterus. Solitary pulmonary metastasis is the most likely diagnosis in a patient with a history of extrapulmonary malignant neoplasm presenting with a pulmonary mass, but it must be distinguished from primary lung cancer [12] in particular of neuroendocrine origin. In our case, distant metastasis to the lungs occurred 5 years after initial treatment which presented as a solitary pulmonary mass.

## Conclusion

Although the incidence of pulmonary metastasis of ENB is extremely rare, it should be kept in mind in patients with ENB presenting with a solitary pulmonary nodule. Because of the occurrence of late distant metastasis, routine imaging surveillance should be considered.

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