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A black adrenal adenoma with high FDG uptake on PET/CT scan in a patient with esophageal carcinoma: A case report

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ABSTRACT

INTRODUCTION: Black adrenal adenoma (BAA) is a rare, benign adrenal lesion with a black or brown appearance. This is the first report of this lesion in a patient with a synchronous esophageal cancer and highlights the importance of considering a false positive finding on a Positron Emission Tomography (PET) scan, which might otherwise preclude resection.

PRESENTATION OF CASE: A 73-year-old male was diagnosed with mid-esophagus carcinoma. Computed tomography scan revealed an enlarged left adrenal gland. Plasma adrenocorticotropic hormones levels were normal. To characterize the adrenal lesion, a PET scan was obtained which showed high uptake of ¹⁸F-fluoro-2-deoxy-D-glucose (FDG), consistent with a metastasis, suggesting T3N2M1, clinical stage IV esophageal cancer. After two courses of neo-adjuvant therapy, sub-total esophagectomy and left adrenalectomy were performed. The adrenal tumor was soft, and black in color, diagnosed as a BAA on histology. The pathologic stage of the esophageal cancer was T3N0M0, Stage II. Six months after surgery, he is alive without recurrence.

DISCUSSION: High FDG uptake by an adrenal lesion on PET scan, as in this patient, usually suggests a metastatic lesion. Although rare, patients with esophageal cancer and adrenal metastases have been reported to have long-term survival, so it is important to characterize an adrenal lesion when found.

CONCLUSION: Most adrenal lesions with high FDG uptake are malignant, but BAA is also positive on PET scan. Although rare, BAA should be considered in patients with solitary adrenal lesions with high uptake on PET scan, even in the presence of a malignancy.

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1. Introduction

Black adrenal adenoma (BAA) is a rare benign adrenal lesion with black or brown appearance containing lipofuscin [1]. BAA has been reported to have a false-positive appearance as a malignancy on Computed Tomography (CT) scan, Magnetic Resonance Imaging (MRI), and Positron Emission Tomography/Computed Tomography (PET/CT) scan with ¹⁸F-fluoro-2-deoxy-D-glucose (FDG) [2]. This is the first report of the PET-CT appearance of a BAA, in the presence of an esophageal cancer, and highlights the importance of considering this rare lesion.

2. Presentation of case

A 73-year-old male was referred with a one month history of dysphagia. He has a 25 pack-year smoking history and drank 40 g of alcohol daily. At the age of 47, he underwent endoscopic treatment of a gastric cancer, and has had hypertension since age 67. He reports a recent weight loss of 8 kg over the last two months. Endoscopy revealed a circumferential tumor of the thoracic esophagus, and biopsy showed squamous cell carcinoma. CT scan revealed an esophageal tumor in the upper to middle thoracic esophagus and enlarged subcarinal lymph nodes (Fig. 1A), main bronchus lymph nodes, right recurrent nerve lymph nodes and an enlarged left adrenal gland (Fig. 1B). Plasma adrenocorticotropic hormones levels were normal. MRI revealed an adrenal tumor with no fat component (Fig. 2A, B). PET/CT scan with FDG (SIMENS Biograph 16, SIMENS, Munich, Germany) showed high accumulation, with a maximum standard uptake value (max SUV) 10.13. The background liver has max SUV 2.75 (Fig. 3). Taken together, these results

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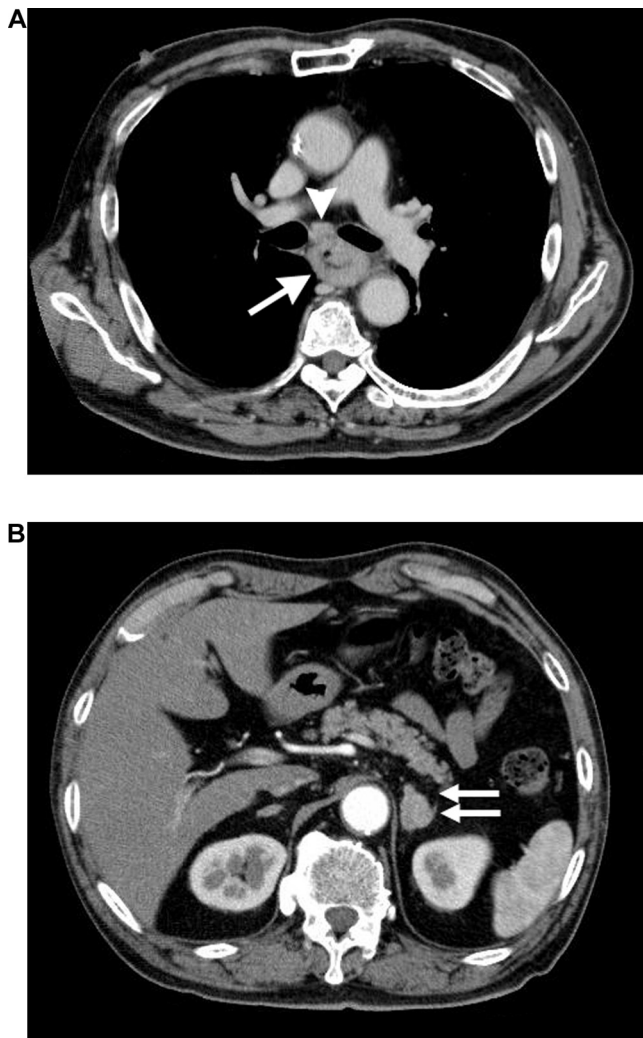


Fig. 1. (A,B) Computed tomography scan revealed an esophageal tumor in the upper to middle thoracic esophagus (arrow) and enlarged subcarinal lymph nodes (arrow head) (1A). The left adrenal gland was enlarged (double arrows), suspected to contain a metastasis from the primary esophageal cancer (1B).



Fig. 2. (A,B) Magnetic Resonance Imaging revealed that there is no difference in signal intensity comparing in-phase (2A) and out-of phase images (2B). The adrenal tumor had no fat component.

suggested that the likelihood of a left adrenal metastasis was high, with a clinical stage T3N2M1, stage IV esophageal cancer.

After two courses of neo-adjuvant FP therapy (5-fluorouracil and cisplatin) as induction chemotherapy, his dysphagia worsened. A CT scan was obtained which showed that the primary tumor and the enlarged regional lymph nodes had decreased in size remarkably, but the adrenal tumor was unchanged. Sub-total esophagectomy with gastric tube reconstruction and left adrenalectomy were performed. The adrenal tumor was soft, and the split surface was black (Fig. 4). Histologically, cells with eosinophilic cytoplasm proliferated monotonically and uniformly, containing brown pigment seen on H&E stained sections, and diagnosed as a BAA (Fig. 5). The esophageal lesions were a mixture of viable cancer cells and foreign body reactions involving many multinuclear giant cells, and it seemed that the proportion of viable cancer cells had reduced to about 1/3 to 2/3 following chemotherapy. There was a foreign body reaction with invasion of multinucleated giant cells in the lymph nodes, with a possibility of metastasis, but there were no tumor cells seen. The final pathological stage was T3N0M0, Stage II. As a result, we believe that the surgery has a reasonable likelihood of being a curative resection. Six months after surgery, he is alive and without recurrence.

3. Discussion

BAAs have been described in resected adrenal specimens in patients with Cushing's syndrome and preclinical Cushing's syndrome, and are rarely diagnosed in incidental adrenal masses [2]. The hypertrophied cortical cells of the black adenoma in the zona reticularis contain the pigment lipofuscin. This pigment accounts for its black or brown appearance [3]. FDG uptake on PET/CT scan by adenomas usually does not exceed that of the liver. The scans are usually interpreted as positive for malignancy if the FDG uptake is equal or greater than that of the liver. A sensitivity of 100% and a specificity of 94% have been reported for adrenal lesions detected by CT or MRI [4]. There are only two reports of PET scan evaluation of BAAs. One study showed a, max SUV 3.6, only slightly higher than the background liver max SUV 3.2 [3]. The other study showed a max SUV 8.3, clearly higher than the background liver max of SUV 2.6 [2]. This report concluded that it is difficult to distinguish malignant adrenal tumors from BAA on CT scan, MRI, or PET/CT. In the present patient, the max SUV was 10.13, clearly higher than the background liver max of SUV 2.75, which confirmed a high level of accumulation with FDG, greater than in previous reports. This contributed to the difficulty in differentiating a BAA from a malignant lesion on the basis of imaging studies.



Fig. 3. The positron emission tomography/computed tomography scan showed a left adrenal gland (arrow) with high uptake of ^{18}F -fluoro-2-deoxy-D-glucose, SUV_{max} 10.13.

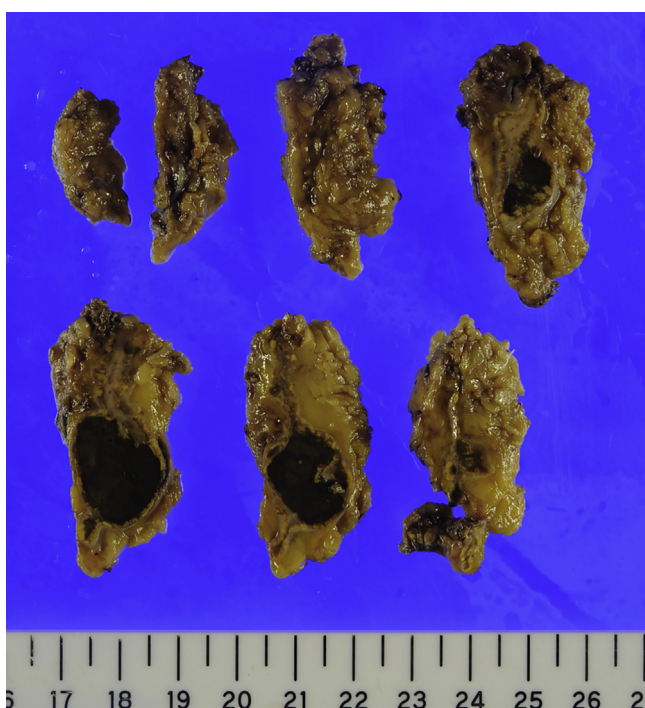


Fig. 4. The left adrenal gland specimen was soft and the split surface black.

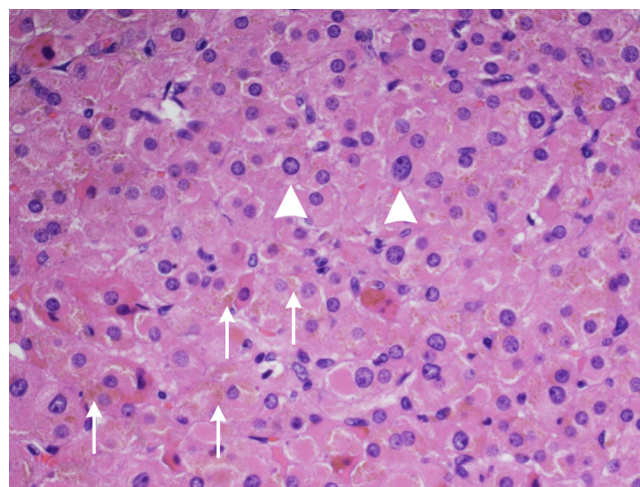


Fig. 5. Histologically, cells with eosinophilic cytoplasm (arrow heads) proliferated monotonically and uniformly, contain brown pigment (arrows) by H&E staining ($\times 400$), and diagnosed as a BAA.

In the present patient, we found high accumulation of FDG on PET scan, and since a fat component was not conspicuous in MRI, the adrenal gland was judged to be more likely to harbor a metastasis than an adenoma. For the diagnosis of malignancy, endoscopic ultrasound fine-needle aspiration of the adrenal is reported to have a sensitivity, specificity, positive predictive value and negative predictive value of 86%, 97%, 96% and 89%, respectively, without serious adverse events [5]. However, we went directly to surgical resection in this patient because of the worsening symptom of dysphagia, necessitating early resection.

Among patients with metastases from carcinoma of the esophagus, lesions are commonly found in the lung (28%), liver (18%), and vertebrae (15%) at autopsy. Metastases to the adrenal glands are found in 10% [6]. In general, patients with metastases to the adrenal gland from esophageal cancer have an extremely poor prognosis. In addition, patients with esophageal cancer rarely have isolated metastases to the adrenal gland, which limits the utility of adrenalectomy in these patients. Two patients have been reported to undergo resection of synchronous metastases to the adrenal gland at the time of esophagectomy [7,8]. To date, we identified reports of five patients who underwent resection of metachronous metastases to the adrenal gland from carcinoma of the esophagus [9–13]. Although the number of patients is small, all survived for more than one year, and one patient had no relapse after 47 months [10].

Dysphagia worsened in the present patient after two courses of chemotherapy, although the esophageal malignancy became smaller. The effect of chemotherapy was considered to be a partial response. The adrenal tumor was unchanged after chemotherapy, suggesting the possibility that it was not a metastasis. In view of the fact that this patient had a synchronous isolated adrenal lesion, the decision was made to resect both the esophageal cancer and the adrenal gland simultaneously.

4. Conclusions

Although most adrenal masses with high FDG uptake on PET scan are malignant, the rare BAA is also positive. PET scan is helpful in the evaluation of patients with esophageal cancer. BAA should be considered in a patient with a solitary adrenal lesion that has high FDG uptake on PET scan, even in the presence of a malignancy.

Conflicts of interest

The authors declare no conflicts of interest.

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None.

Ethical approval

Review of this case report was waived by the Jichi Medical University Institutional Review Board.

Consent

Written and signed consent was given by the patient for this case report.

Author contribution

Shiro Matsumoto: Conception of Study, Acquisition and analysis of data, drafting and critical revisions of the article, approval of final version

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This case report conforms to the SCARE criteria [14].

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