

Retroperitoneal Bronchogenic Cyst Presenting Paraadrenal Tumor Incidentally Detected by ^{18}F -FDG PET/CT

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Abstract A follow-up ^{18}F -fluorodeoxyglucose (^{18}F -FDG) PET/CT scan of a 57-year-old asymptomatic male who had undergone total thyroidectomy for thyroid cancer revealed a 5.0×4.0 -cm, well-defined, ovoid-shaped mass around the left adrenal gland without definite FDG uptake. On the adrenal CT scan, the left paraadrenal tumor showed high attenuation on the precontrast scan without enhancement. The average Hounsfield unit (HU) was 58.1 on the precontrast scan and 58.4 on the postcontrast scan. The patient underwent laparoscopic adrenalectomy for resection of the left paraadrenal tumor. The final histopathologic examination revealed a bronchogenic cyst. Although retroperitoneal bronchogenic cysts

are rare, they should be considered in the differential diagnosis of retroperitoneal cystic tumors. The preoperative diagnosis is difficult, but a contrast-enhanced CT scan or ^{18}F -FDG PET/CT scan may be useful for differentiating hyperattenuated cysts from other soft tissue masses.

Keywords Bronchogenic cyst · Retroperitoneal cyst · CT · ^{18}F -FDG PET/CT

Introduction

Bronchogenic cysts are congenital foregut malformations that arise from an abnormal budding of the tracheobronchial tree [1, 2]. The usual location of bronchogenic cysts is the tracheobronchial tree, esophagus, or mediastinum. However, they may occur in other unusual locations such as the subdiaphragmatic area. Retroperitoneal bronchogenic cysts are rare. While CT scan findings of retroperitoneal bronchogenic cysts have previously been reported, few data address ^{18}F -FDG PET/CT scan findings with regard to retroperitoneal bronchogenic cysts. We report a rare case of paraadrenal bronchogenic cyst detected by an ^{18}F -FDG PET/CT scan.

Case Report

A 57-year-old asymptomatic male had undergone a follow-up ^{18}F -FDG PET/CT scan for a total thyroidectomy 15 years previously because of papillary thyroid cancer. The ^{18}F -FDG PET/CT scan revealed a 5.0×4.0 -cm, well-defined, ovoid-shaped mass around the left adrenal gland without definite FDG uptake and a 1-cm nodular lesion in the right adrenal gland with FDG uptake [maximum standardized uptake value (SUVmax) 2.5] (Fig. 1). On the adrenal CT scan, the left paraadrenal tumor showed slightly high attenuation on the

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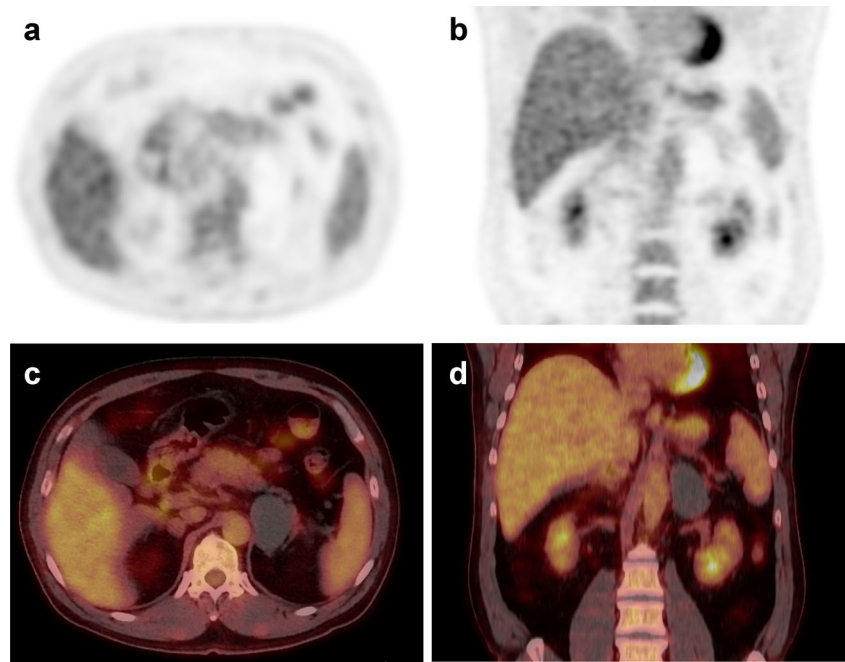
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Fig. 1 PET axial (a), PET coronal (b), fused axial (c), and fused coronal (d) ^{18}F -FDG PET/CT revealed almost no FDG uptake of the left paraadrenal cystic tumor and mild FDG uptake of the right adrenal tumor (SUVmax 2.5)



precontrast scan without enhancement (average HU, 58.1 on precontrast scan and 58.4 on postcontrast scan). The average HU of the right adrenal tumor was 29.6 on the precontrast scan and 77.9 on the postcontrast scan (Fig. 2). Laboratory data including metanephrine, normetanephrine, norepinephrine, vanillylmandelic acid, cortisol, and aldosterone levels were all within normal range. Because the size of the left paraadrenal tumor was greater than 4 cm, the patient underwent laparoscopic adrenalectomy. The tumor was a cystic lesion measuring 4.8×2.5 cm in size and located superior to the left adrenal gland (Fig. 3). Histopathologic examination reported that the cyst was lined with pseudostratified ciliated columnar epithelium (Fig. 4). This finding was consistent with a bronchogenic cyst. Because CT findings of the right adrenal

gland tumor were consistent with adrenal adenoma, the lesion was not removed. The patient had no postoperative complications and was discharged home 8 days after the operation.

Discussion

Bronchogenic cysts are congenital foregut malformations that arise from an abnormal budding of the tracheobronchial tree during the 3rd to 7th week of development. If attachment to the primitive foregut persists, the cyst is usually located in the tracheobronchial tree, esophagus, or mediastinum. In particular, the posterior part of the carina is a common site, while the retroperitoneal area is rare. Although the exact mechanism for

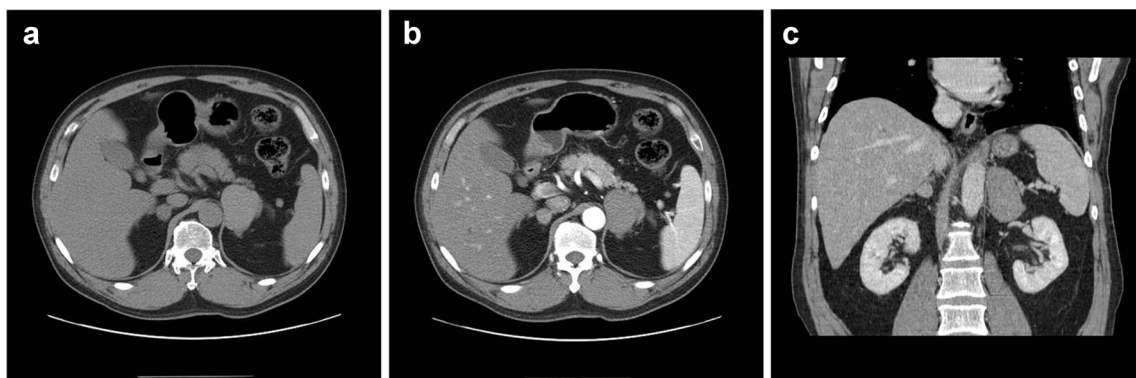


Fig. 2 Axial precontrast (a), postcontrast (b), and coronal postcontrast (c) CT scan revealed a 5-cm, ovoid-shaped, well-defined, high-attenuation left paraadrenal mass without enhancement and 1-cm-sized enhancing nodule in the right adrenal gland

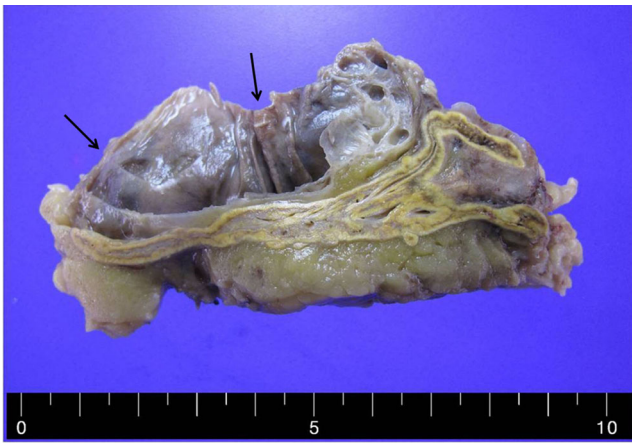


Fig. 3 Gross features of an opened left retroperitoneal cyst (arrows) and left adrenal gland. A 4.8×2.5-cm cyst was located superior to left adrenal gland

formation of retroperitoneal bronchogenic cysts is unknown, modern theory suggests the thoracic and abdominal cavities are connected by the pericardio-peritoneal canal in early embryogenesis [1, 2]. When the canal is later divided by fusion of the pleuroperitoneal membranes, a portion of the tracheobronchial tree could be pinched off and migrate, resulting in the formation of a retroperitoneal bronchogenic cyst. Retroperitoneal bronchogenic cysts can occur in a wide age range and are found equally in males and females. Among all cases of retroperitoneal bronchogenic cysts reported in the literature, 82 % of cases occur in the left side of the retroperitoneal region, as in our case [3]. Histopathologically confirmed bronchogenic cysts are usually unilocular and have thin walls lined by ciliated respiratory epithelium with bronchial glands, nerves, and cartilage [1, 2].

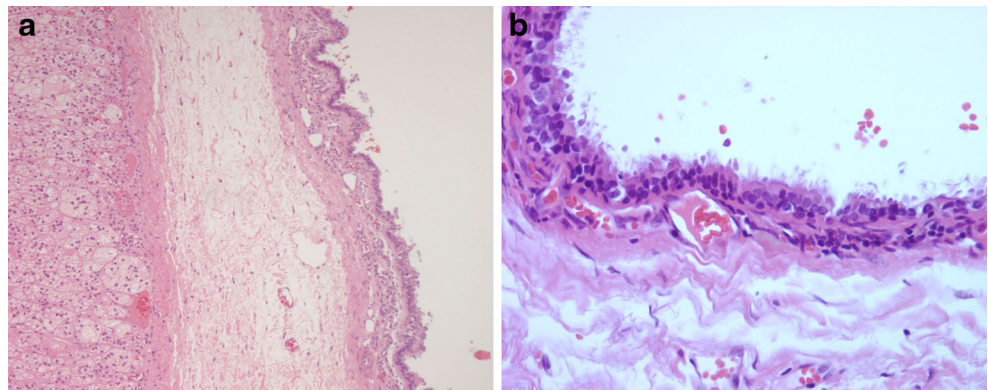
Surgical excision is recommended for retroperitoneal bronchogenic cysts to establish a definitive diagnosis. Although most patients are asymptomatic, these lesions become secondarily infected, perforated, or large enough to have a mass effect on adjacent organs. Sometimes, they present symptoms such as flank or abdominal pain. Thus,

excision is the only method to confirm the diagnosis, alleviate symptoms, and prevent complications such as infection and risk of malignancy. Our present case showed no definite clinical symptoms or laboratory abnormalities. Due to the tumor's large size (approximately 5 cm on the CT image), the patient underwent laparoscopic excision for accurate diagnosis [1, 2].

The differential diagnosis of a retroperitoneal cystic mass includes lymphangioma, mucinous cystadenoma, cystic teratoma, cystic mesothelioma, cystic metastases, cysts of urogenital and foregut origin, such as Müllerian cysts or bronchogenic cysts, pancreatic pseudocysts, lymphoceles, urinoma, and hematoma [4]. Although there is substantial overlap of image findings in various retroperitoneal cysts, some image findings may provide information of bronchogenic cysts. On CT scans, bronchogenic cysts are typically rounded, well circumscribed, and hypoattenuated without enhancement [5, 6]. Sometimes, bronchogenic cysts could appear as a hyperattenuated homogeneous mass. Fluid contents within bronchogenic cysts range from a watery liquid to hemorrhagic fluid to a very viscous proteinaceous mucoid material. Thus, bronchogenic cysts could present as a variable attenuated cystic mass lesion on CT scans with variable signal intensity on MR images because of its content variability. Based on cyst fluid analysis, it has been reported that calcium is a major factor contributing to high attenuation on CT scans. MR imaging can be a useful modality for differentiating a highly attenuating cyst from soft tissue masses on CT scans. On MR images, bronchogenic cysts are typically isointense or hyperintense to cerebrospinal fluid (CSF) on T2-weighted images [6]. A lesion that is hypointense to CSF on T2-weighted images should be considered with caution.

To our knowledge, there are few published reports on ^{18}F -FDG PET/CT findings of bronchogenic cysts. Huang et al. reported a case of retroperitoneal uncomplicated bronchogenic cysts with no FDG uptake on ^{18}F -FDG PET/CT scans [7]. Yekeler et al. also reported a case of an intramural esophageal bronchogenic cyst without FDG uptake on an ^{18}F -FDG PET/

Fig. 4 Histologic section of the left retroperitoneal cyst with hematoxylin and eosin staining: ×100 (a); ×400 (b). The cyst was lined by pseudostratified ciliated columnar epithelium, indicating the bronchogenic cyst



CT scan [8]. This case also showed a paraadrenal tumor without definite FDG uptake in the left paraadrenal area. Although malignant transformation of bronchogenic cysts is extremely rare, there are several reports of malignancy arising in bronchogenic cysts [9–12]. Tsai et al. reviewed 22 cases of bronchogenic cyst with malignant change and summarized the clinical information, outcome, and malignant subtypes [9]. Malignancies arising from bronchogenic cysts include adenocarcinoma, squamous cell carcinoma, anaplastic carcinoma, carcinoid, mucoepidermoid carcinoma, and various types of sarcomas in their review. Aydogdu et al. described a rare case of schwannoma in an intrapulmonary bronchogenic cystic wall with FDG uptake of SUVmax 2.5 [12]. The specimen from our case revealed pseudostratified ciliated columnar epithelium without malignant or inflammatory cells. Since FDG accumulation occurs in malignant and inflammatory tissues, ^{18}F -FDG PET/CT may help detect complications such as infection or malignant change.

Despite the rarity of this pathologic entity, a bronchogenic cyst should be considered in the differential diagnosis of retroperitoneal cystic tumors. Since uncomplicated hyperattenuated cysts such as retroperitoneal bronchogenic cysts show almost no FDG uptake, FDG PET/CT may be useful for differentiating hyperattenuated cysts from other soft tissue masses and evaluating complications such as infection or malignant change.

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Informed Consent All procedures followed were performed in accordance with the ethical standards of the responsible committee on human experimentation and with the Helsinki Declaration of 1975, as revised in 2000. The study design and exemption of informed consent were approved by the Institutional Review Board of the Soonchunhyang University Cheonan Hospital.

Conflict of Interest Ye Ri Yoon, Jiyoun Choi, Sang Mi Lee, Yeo Joo Kim, Hyun Deuk Cho, Jeong Won Lee, and Youn Soo Jeon declare that they have no conflict of interest.

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