
Case Report

Poorly Differentiated Adenocarcinoma Arising from a Cervical Bronchial Cyst

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Bronchogenic cysts with malignant change are rarely reported. We describe a case of poorly differentiated adenocarcinoma arising from a cervical bronchial cyst in a patient presenting with a thyroid mass, cervical lymphadenopathy, and initial biopsy suggestive of papillary thyroid carcinoma. The clinical presentation, intraoperative findings, radiographic images, and pathology slides are presented. To our knowledge, this is the first report of a poorly differentiated adenocarcinoma arising from a bronchial cyst in the cervical region.

Key Words: Head, neck.

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INTRODUCTION

Bronchogenic cysts are a rare, benign congenital malformation derived from the embryonic foregut and occur most commonly in the mediastinum in close proximity to the thoracic trachea. Ectopic locations in the neck are rare and have been described mostly in the pediatric population.¹ In adults, cervical bronchial cysts have been reported in the paratracheal, cutaneous, lingual, and supraclavicular areas.² Malignant transformation of a bronchial cyst arising in the neck has never been reported. We present the first case of a poorly differentiated adenocarcinoma arising from a cervical bronchial cyst.

CASE REPORT

A 32-year-old female presented with a 3-month history of throat pain, an enlarging left neck mass, and left vocal cord paresis. CT imaging showed a 4.2 × 3.5 cm

mass arising from the left thyroid lobe with left cervical lymphadenopathy. Ultrasound-guided fine-needle aspiration of the left thyroid mass was suggestive of papillary thyroid carcinoma (Fig. 1a).

Based on the presumed diagnosis of a well-differentiated thyroid carcinoma, the patient was taken to the operating room for a planned total thyroidectomy and left neck dissection. Intraoperatively, the patient was found to have a large left thyroid mass with gross tracheoesophageal invasion and extensive jugular chain lymphadenopathy densely adherent to the common carotid artery. Intraoperative frozen specimen analysis showed a poorly differentiated adenocarcinoma. Subtotal thyroidectomy with a limited neck dissection was performed.

Postoperative PET/CT imaging 3 weeks later showed a hypermetabolic mass in the area of the left thyroid lobe, left neck (levels II and IV) and tracheoesophageal invasion (Fig. 2a), consistent with diffuse aggressive disease encountered intraoperatively. Additionally, the patient had diffuse substernal and pulmonary metastases (Fig. 2b).

Microscopically, a poorly differentiated adenocarcinoma was seen extensively invading into the thyroid gland (Fig. 3a), skeletal muscle, and fibroadipose tissue. Furthermore, a cystic structure was present, lined by ciliated columnar epithelium with a single underlying smooth muscle layer with dysplastic changes (Fig. 3b); focally, the cyst lining was seen to transition into invasive adenocarcinoma and contained mucin. Extensive immunohistochemistry was performed; the tumor cells were strongly positive for CK7, CEA, and P53 and moderately positive for CA19.9 and vimentin. Tumor cells

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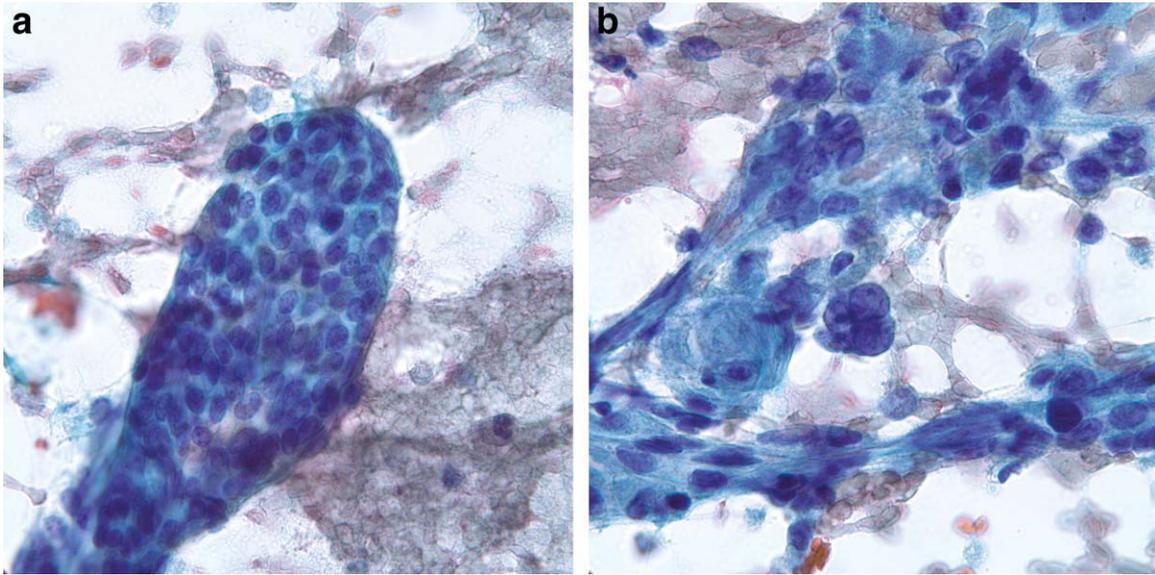


Fig. 1. Fine-needle aspiration (FNA) of left thyroid mass. (a) This portion of the FNA shows subtle papillary and acinar architecture with clusters of atypical epithelioid cells. Cells show round to oval nuclei with variable nuclear pleomorphism and some nuclear grooving, originally read as papillary thyroid carcinoma. (b) Different portion of FNA showing more poorly differentiated cells; retrospective pathology addendum was consistent with a poorly differentiated carcinoma. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

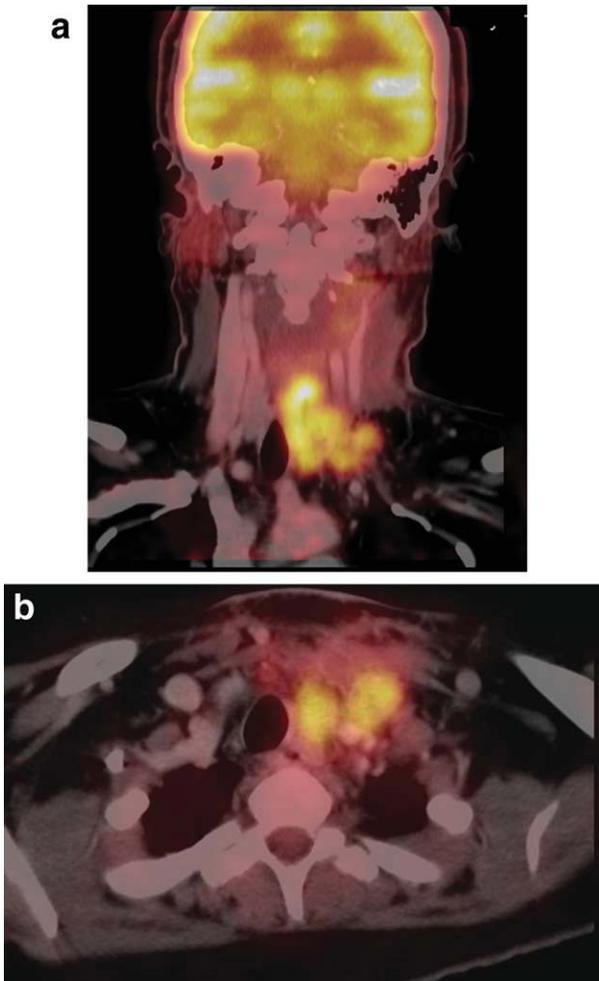


Fig. 2. PET-CT scans obtained postoperatively. (a) Coronal PET-CT showing an enhancing mass originating from the left lobe of the thyroid with extensive ipsilateral cervical adenopathy involving nodal stations II-IV. (b) Axial PET-CT showing substernal extension of the mass along with mediastinal adenopathy. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

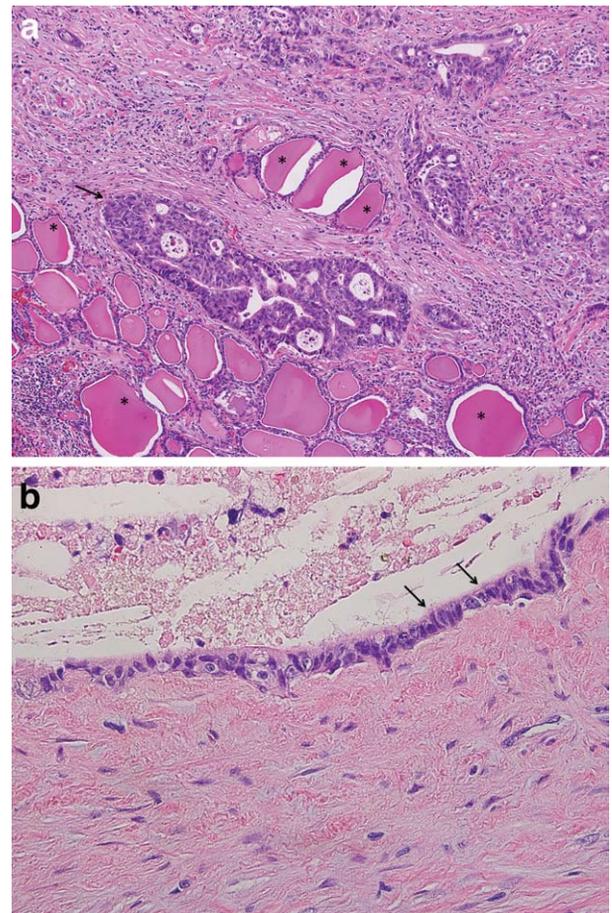


Fig. 3. Histopathology of left thyroid mass. (a) Thyroid parenchyma with infiltrating malignant glands consistent with adenocarcinoma (arrow). Some thyroid follicles indicated by *. (b) Epithelial lining of cervical bronchial cyst showing dysplastic changes. Note the presence of residual cilia and terminal bar (arrow). Other areas demonstrate full thickness adenocarcinoma in situ (not shown here). [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

were negative for CK20, TTF-1, TG, GCDFP15, ER, PR, HER2, Mammaglobin, CA125, WT-1, PAX-8, Calretinin, and keratin 5/6. The histopathology was most compatible with a poorly differentiated adenocarcinoma arising from a cervical bronchogenic cyst (detailed histopathologic review will be reported separately).

Retrospective review of the preoperative fine-needle aspiration showed some clusters of atypical epithelioid cells suggestive of a more poorly differentiated carcinoma despite subtle papillary and acinar architecture (Fig. 1b). The patient is undergoing concurrent chemoradiation with taxol and carboplatin.

DISCUSSION

Bronchogenic cysts in the cervical region are usually diagnosed in the pediatric population as asymptomatic cervical masses.¹ They are thought to be derived from abnormal sequestrations or budding of the tracheal primordia. These are pinched off at the time of fusion of the mesenchymal bars of the sternum, which leaves respiratory tissue outside the thorax.³

Cervical bronchial cysts in adults are rare, with only 16 reported cases. These were all benign and presented most often as an asymptomatic neck mass. In a review by Moz et al.,⁴ the thyroid and paratracheal region were most frequently affected, and the majority of cysts were midline. The proposed treatment of choice for bronchial cysts arising from any location is surgical excision to prevent the complications of infection, rupture, compressive symptoms, and malignant degeneration. Malignancies arising from bronchial cysts have been shown primarily in intrathoracic cysts. Reported malignancies arising from thoracic bronchial cysts include adenocarcinoma, squamous cell carcinoma, anaplastic carcinoma, and various types of sarcomas.

In contrast to thoracic bronchial cysts, carcinoma arising from bronchial cysts above the clavicle has been definitively reported only once. Tanita et al.³ described a case of malignant melanoma arising from a cutaneous bronchogenic cyst in the scapular area. Mizukami et al.⁵ reported a mucoepidermoid carcinoma of the thyroid gland in a 44-year-old female and speculated possible bronchial cyst origin among other hypotheses. A cystic wall demonstrating malignant change, however, was not seen. To our knowledge, our patient represents the first reported case of poorly differentiated adenocarcinoma arising from cervical bronchial cyst origin.

Additionally, the clinical presentation of this patient is worthy of discussion. As a head and neck surgeon, patients with thyroid masses and concomitant lymphadenopathy are commonly encountered. This patient presented with what appeared to be a well-differentiated thyroid carcinoma. She had a neck mass, lateral cervical

lymphadenopathy, vocal cord paresis, and a preoperative ultrasound-guided needle biopsy suggesting papillary thyroid carcinoma. Intraoperatively, suspicion of a more aggressive etiology arose due to extensive unresectable tumor. The more likely differential diagnoses of aggressive thyroid tumors, including anaplastic carcinoma and metastatic adenocarcinoma from the breast, colon, and ovary, were eliminated with immunostaining. Instead, the tumor was found to arise from a cystic structure consistent with bronchogenic origin on immunostaining.

Retrospective analysis of the preoperative needle biopsy did in fact show evidence of poor differentiation (Fig. 1b). It is unclear why there were areas in the FNA consistent with papillary thyroid carcinoma, as there was no evidence of well-differentiated thyroid carcinoma in the surgical specimen. One possible explanation is that papillary architecture can be seen in inflammatory thyroid disease, such as Hashimoto's thyroiditis. This case emphasizes the importance of clinical suspicion despite fine-needle aspiration findings, which can be misinterpreted due to the small amount of specimen collected. The clinical presentation of pain, neck mass, and vocal cord paresis, together with intraoperative findings of extensive, unresectable disease correlated with the histopathology of a poorly differentiated carcinoma.

This is the first report of a poorly differentiated adenocarcinoma arising from a bronchogenic cyst in the cervical region and highlights the importance of having a clinical suspicion for a more poorly differentiated process in patients presenting with pain, vocal cord paresis, and extensive radiographic findings.

CONCLUSION

Malignancies arising from cervical bronchial cysts are exceedingly rare. We present the unique case of a patient with a thyroid mass and a needle aspirate showing well-differentiated thyroid carcinoma that was subsequently diagnosed as a poorly differentiated adenocarcinoma from cervical bronchial cyst origin.

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