PRIMARY ADENOCARCINOMA IN THE SETTING OF A BRANCHIAL CLEFT CYST

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ABSTRACT

Objectives: Present a case of adenocarcinoma in the setting of a branchial cleft cyst. Discuss the differential of primary branchial cleft cyst adenocarcinoma vs. undiscovered primary with a rare metastasis to the branchial cleft. Review the literature of branchial cleft cyst carcinomas, all of which are squamous cell carcinomas.

Methods: Chart review of the patient was performed including clinical course, PET imaging and pathology. A PubMed literature review was performed.

Results: Our patient is a 58-year-old male who underwent excision of a cystic mass, and was found to have a focus of adenocarcinoma within the setting of a branchial cleft cyst. Although the patient had two other primary cancers, metastatic melanoma and follicular lymphoma, an alternative primary source for his branchial cleft cyst carcinoma was not found by PET/CT imaging. No other cases of adenocarcinomas in the setting of a branchial cleft cyst were found in literature review. Branchial cleft cysts can contain rests of salivary tissue, which may be the source for the adenocarcinoma.

Conclusions: The existence of primary carcinomas of the branchial cleft cyst are controversial in the literature. As primary carcinomas of the head and neck are often difficult to detect. This case may represent either primary or metastatic adenocarcinoma in the branchial cleft cyst. Primary adenocarcinoma may arise from an embryologic rest of glandular tissue in the branchial cleft cyst. Metastases of adenocarcinoma are more likely to involve lymph node basins than branchial cleft cysts, with the latter being previously unreported.

INTRODUCTION

• The embryologic branchial apparatus is responsible for the formation of the fetal face, pharynx, and neck structures 1
• Structures which compose the embryologic branchial apparatus can fail to obliterate, leaving cystic neck masses, neck fistulae, sinuses tracts, or cartilaginous elements in the adult neck 2
• Branchial cleft cysts are generally benign and can be treated with complete surgical excision
• The diagnosis of primary branchial cleft cyst carcinoma is controversial, with some considering these cases misdiagnoses of cystic cervical metastasis of squamous cell carcinoma. Regardless, there are well-defined diagnostic criteria for primary branchiogenic carcinoma, with one requirement being squamous cell carcinoma 3
• The only reported primary branchial cleft carcinomas prior to this case in the English-language literature have been squamous cell carcinoma
• Generally, cases of metastatic adenocarcinoma in the neck are located in the cervical lymph nodes
• Branchial cleft cysts have been reported to contain ectopic salivary tissue 4

METHODS

• Case report of a single patient
• Chart review including clinical course, operations, pathologic findings, and imaging results
• Staining for mixed cytokeratin, CK7, CK20, and TTF-1 were performed on the specimen
• An English-language PubMed literature review was performed using the keywords “branchial cleft cyst carcinoma,” “primary cervical carcinoma,” “primary branchiogenic carcinoma,” “cystic neck carcinoma,” “cystic cervical carcinoma,” “BCC squamous cell carcinoma,” “salivary heterotopia to the neck,” “adenocarcinoma branchial cleft cyst”

FIGURE

CASE REPORT

• 58 year old male with right neck mass incidentally found on PET/CT performed for malignant melanoma
• Past medical history: malignant melanoma and follicular lymphoma of right parotid
• Further imaging was performed, including CT neck (see Figure, Panel A) and US
• Mass was surgically excised, with uneventful post-operative course
• Pathology (Panel B-D) showed cystic mass with benign non-keratinizing squamous epithelium and benign respiratory epithelium, consistent with branchial cleft cyst. Pathology also showed glandular proliferation immediately adjacent to the cyst, with enlarged vesicular nuclei, distinct nucleoli, and moderate eosinophilic cytoplasm. Stratification and loss of polarity, areas of confluence, and cribriforming glands supports a diagnosis of adenocarcinoma. The tumor cells are positive for pancytokeratin and are negative for CK7, CK20, and TTF by immunohistochemistry with appropriate controls. Negative staining for TTF-1 makes the diagnosis of primary thyroid adenocarcinoma unlikely.
• Follow-up PET/CT imaging revealed no primary source of an adenocarcinoma

DISCUSSION

• Branchiogenic carcinoma is defined as squamous cell carcinoma within a branchial cleft cyst
• No other reported cases of non-thyroid adenocarcinoma within a branchial cleft cyst were found on literature review
• Three possibilities include primary adenocarcinoma (possibly arising from ectopic salivary tissue or metaplasia), metastatic non-thyroid adenocarcinoma in an unusual location, or thyroid carcinoma (though TTF-1 negative staining makes this unlikely).
• To our knowledge, primary adenocarcinoma in a branchial cleft cyst has never before been reported, which makes this the first possible case

REFERENCES