Case Report

ISSN (0):2395-2822; ISSN (P):2395-2814

A Rare Case of Primary Papillary Carcinoma of Ectopic Thyroid in a Branchial Cleft Cyst.

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Received: January 2017 Accepted: January 2017

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ABSTRACT

Cysts arising from the lateral neck commonly represent benign lesions such as branchial cysts and lymphangiomas. Occasionally a cyst of the lateral cervical region is a malignant lesion metastatic from a papillary thyroid carcinoma. Ectopic thyroid tissue within a branchial cleft cyst may present as an exceptional finding: a primary thyroid carcinoma. Here we a report a rare case of ectopic thyroid papillary carcinoma that presented as lateral neck cyst with preoperative diagnosis of branchial cleft cyst. After complete surgical resection and histopathologic examination, the lesion was determined to be a branchial cleft cyst with primary thyroid papillary carcinoma.

Keywords: ectopic thyroid, branchial cleft cyst, papillary carcinoma, surgical resection, histopathological diagnosis.

INTRODUCTION

Branchial cleft cysts are the most common lateral cystic neck masses. [1] Ectopic thyroid tissue within a branchial cleft cyst is a rare phenomenon, and papillary thyroid carcinoma arising from this tissue is extremely rare. [2] We report a case of incidentally found primary papillary thyroid carcinoma in a branchial cleft cyst. Although most lateral cystic masses of neck in young adult patients are found to be benign, an occult primary papillary carcinoma of thyroid within a branchial cleft cyst should be considered in the differential diagnosis. Our objective is to draw the reader's attention to this possibility of an unexpected primary papillary carcinoma in an ectopic thyroid tissue within the branchial cleft cyst.

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CASE REPORT

A 24 year old lady presented with history of right lateral neck swelling since 5 months with progressive increase in size and difficulty in breathing on attaining supine position. The past medical history of this lady was otherwise unremarkable without any previous neck radiation

exposure. Physical examination showed a 4.5 x 6.5 cm smooth, round, non-tender mass at the anterior border of the middle third of the right sternocleidomastoid muscle. CECT revealed a well defined, non-enhancing cystic lesion posterior to the sternocleidomastoid with an eccentric focus of enhancement suggesting a diagnosis of third branchial cleft cyst with possibility of malignancy. However, thyroid gland was unremarkable. Bilateral level III, IV and V lymphadenopathy was noted. She was admitted for further treatment. At surgical resection, no tract was discovered connecting the cyst to the pharynx or the hyoid bone and a benign, clear fluid-filled lesion was excised with no signs suggesting invasion. Histopathological diagnosis was consistent with a branchial cyst with a well differentiated papillary thyroid carcinoma. Physical examination, thyroid ultrasound and CT of the neck obtained before surgery showed no evidence of thyroid gland tumor. Total thyroidectomy and selective neck dissection was performed at a later stage. However, histopathology of thyroid revealed no signs of malignancy.

DISCUSSION

The thyroid gland originates from the primitive alimentary tract and is predominantly of endodermal origin. It arises as a midline diverticulum from the floor of the pharynx in the region of the foramen cecum at about the third

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gestational week and becomes recognizable about 1 month after conception. The main body of the thyroid descends into the neck from its origin and migrates caudally. It then develops into a bilobated solid organ. With the resorption of the thyroglossal duct, the original attachment to the buccal cavity at the foramen cecum, around the sixth week of gestation the developing gland reaches its final location. [3]

According to Weller and more recently Hoyes, Kershaw and Williams follicular cells from the fourth branchial pouch contribute to development of the thyroid gland lobes. A failed obliteration of 2nd, 3rd and 4th branchial pouch was thought caused to be the etiology of branchial cysts. [4] This theory has been abandoned and it is now believed that epithelial cell inclusions from the upper airway and GI tract enter the lymphatics and reach the lateral neck lymph nodes. This "cystic colonization would promote a degeneration" of lymph nodes. It has been postulated that similar changes might occur if aberrant thyroid tissue colonizes the lateral neck lymph node as a result of an "embryologic accident".[5]

Ectopic thyroid tissue is reported in 7% of adults and is frequently found along the course of thyroglossal duct or around the two lobes of the gland. [6] Other possible sides of ectopic localization are anterior tongue, larynx, trachea, esophagus, mediastinum, pericardium, diaphragm and rarely, neck branchial cyst. [7] Previous reports showed that ectopic thyroid tissue may present metastasis from thyroid carcinoma, and very rarely it may harbour a primary thyroid carcinoma. Of the latter, about one hundred cases have been so far described in literature. [8] Most of them have been shown to occur in the thyroglossal duct, 1% out of all thyroglossal cysts carcinomas are papillary carcinomas. [9]

The initial assessment of a patient who presents with a neck mass should include a thorough evaluation of the head and neck, a survey of risk factors for malignancy, an FNAC analysis and imaging with ultrasound or CT. The presentation of a thyroid papillary carcinoma as a branchial cleft cyst has been reported in three situations: cystic lymph node metastasis of thyroid papillary carcinoma, true branchial cleft cyst with concurrent lymph node metastasis of thyroid papillary carcinoma and most rarely ectopic thyroid papillary carcinoma within a branchial cleft cyst. [10]

Regardless of the etiology, the prognosis of patients with carcinoma in lateral neck cysts without a primary site identified after total thyroidectomy appears to be good. This might suggest that removing the cyst is therapeutic if it represents de novo carcinoma in ectopic thyroid tissue. It could also represent a missed primary in the thyroid secondary to a missed micro-carcinoma, and total

thyroidectomy is strongly recommended and selective neck dissection should be considered. Due to the possibility of papillary micro-carcinoma, serial thin sections of all blocks of the totally embedded thyroid should be performed. Adjuvant radioactive iodine might be considered if residual thyroid tissue or disease is suspected.

CONCLUSION

In a lateral cystic neck mass, although rare, occurrence of ectopic thyroid tissue and presence of a papillary thyroid carcinoma is a possibility. Diagnostic ultrasound and CECT of the neck may display the lesion, however histopathological examination is a must for the diagnosis. If FNAC guided ultrasound of the cyst is inconclusive, excisional biopsy is essential to rule out malignancy and to avoid the delay in the diagnosis and management of the unexpected primary papillary carcinoma of ectopic thyroid in a branchial cleft cyst.

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How to cite this article: Pawan KV, Ravikanth R, Sarkar PS, Kumar N, Philip B. A Rare Case of Primary Papillary Carcinoma of Ectopic Thyroid in a Branchial Cleft Cyst. Ann. Int. Med. Den. Res. 2017; 3(2):RD24-RD25.

Source of Support: Nil, Conflict of Interest: None declared