Papillary Thyroid Carcinoma Occurring in Branchial Cleft Cyst: Case Report and Review of Literature

Swei H. Tsung

ABSTRACT

Ectopic thyroid tissue is ubiquitous in the body, most frequently seen at the base of tongue, accounting for approximately 95% of reported cases. Branchial cleft cyst, a congenital abnormality, can occasionally harbor thyroid tissue inside the cyst. These ectopic thyroid tissues can develop the same disease as the orthodox thyroid gland, including the malignant diseases. Papillary thyroid carcinoma arising in the branchial cleft cyst was extremely rare, total of only 14 cases were reported in the literature. Herein, I report additional case which was discovered incidentally after surgery.

Keywords: Branchial cleft cyst, Papillary thyroid carcinoma, Ectopic thyroid tissue, Total thyroidectomy, Radical neck dissection. Radioactive iodine therapy.

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Swei H. Tsung *

M.D.

Department of Pathology, St Mary's Hospital, Loudong, Yilan, Taiwan.

(e-mail: tsung.sweihsung@gmail.com)

**Corresponding Author*

I. INTRODUCTION

From embryology point of view, the thyroid gland originated from the endodermal epithelium near foramen caecum, and descent to the neck in front of hyoid bone. By definition, any thyroid tissue not located in its normal position is called ectopic thyroid tissue (ECT). Noussios et al. [1] published an extensive review on ETC and pointed out that ECT could be ubiquitous in the body, with the most common location being at the base of the tongue, accounting for about 95% of the reported cases. One place that review article failed to mention was the branchial cleft cyst (BCC) which is a congenital birth defect that occurs early in embryonic development. These ectopic thyroid tissues can develop the same diseases as the orthodox thyroid gland, including the malignant diseases [2]. In 1992 Balasurmanniam et al. [3] reported the first case of papillary thyroid carcinoma arising in ectopic thyroid tissue within BCC. To my best knowledge, there were only 14 cases reported in the literature. Herein, I report additional case which was incidentally discovered after surgery.

II. CASE REPORT

A 55 year-old man with a history of hypertension and hyperlipidemia visited the endocrinology outpatient clinic for routine check-up on June 9, 2020. He was told that he had a lump at his right lateral neck. A complete examination of the ear, nose, throat failed to review any other abnormalities. The laboratory data including thyroid function tests were within normal limits. Neck ultrasound evaluation displayed a mixed solid and cystic lesion (Fig. 1 a). Computed tomography (CT) showed a low density cystic lesion (Fig. 1 b). Fine needle aspiration biopsy (FNAB) was performed, the smears

revealed papillary thyroid carcinoma (Fig. 2 a, 2 b, and 2 c). He was thought to have papillary carcinoma of the thyroid with lymph node metastasis. A surgeon was consulted, and total thyroidectomy with lymph node dissection were performed on October 24, 2020. The surgical recovery was uneventful and was then referred to a medical center to evaluate the possible need of radioactive iodine therapy.

Fig. 1. a. Neck ultrasound evaluation showed a mixed solid and cystic lesions 2.5×2.0cm. b. Computed tomography with contrast showed a low density cystic lesion with lymphadenopathy.

III. CYTOPATHOLOGICAL FINDINGS

Among the specimen removed from the neck dissection, a large cystic nodule measuring 2.5×2.0 cm was found. The cyst was lined by a thin squamous epithelium, beneath it are aggregates of lymphocytes, characteristic of BCC (Fig. 2 d). ETC was found within the cyst, from where papillary thyroid

carcinoma developed (Fig. 2 e and 2 f). Associated with BCC, four lymph nodes containing metastatic papillary carcinoma were found. Meticulous sections were done on the whole thyroid gland, one 0.2 cm solid tumor was demonstrated in the right lobe of thyroid gland. The morphology was characteristics of follicular variant of papillary thyroid carcinoma (Fig. 2 e, 2 h).

Fig 2. (a) Fine needle aspiration smears showed a papillary structure. Pap stain x400.

(b) The smears showed hypercellularity, nuclear pleomorphism and hyperchromatism. Pap stain x400.

(c) The malignant cells displayed enlarged nuclei, intranuclear cytoplasmic inclusions, nuclear grooving and powdery chromatin, diagnostic for papillary thyroid carcinoma. Pap stain x1000.

(d) A branchial cleft cyst surrounded by lymphoid tissue and overlaid by a thin squamous epithelium. Adjacent to the lymphoid tissue was ectopic thyroid tissue displaying nodular goiter. H&E stain x400.

(e) Papillary thyroid carcinoma developed in the branchial cyst. H&E stain x200.

(f) High magnification of papillae lined by malignant cells with nuclear clearing, characteristic for papillary thyroid carcinoma. H&E stain x400.

(g) A small follicular variant of thyroid carcinoma in the right lobe of the thyroid. 0.2 cm in diameter. H&E stain x200.

(h) High power view displaying nuclear characteristics similar to that of papillary thyroid carcinoma. H&E stain x400.

IV. DISCUSSION

In present case, although an occult thyroid carcinoma was found, the morphology of which was a variant of papillary thyroid carcinoma, different from that seen in BCC. Therefore, that the occult variant of the papillary thyroid carcinoma was culprit spreading to BCC and the lymph nodes is unlikely.

The papillary thyroid carcinoma arising BCC is extremely rare. Sagit et al. reviewed the literature in 2013 and found only 11 cases including their own [4]. In 2019, Hong et al, reported additional three cases [5]. Up to the present time, a

total of 15 cases in the literature including the present case were summarized in Table 1. Preoperatively, it is difficult to diagnose BCC harboring papillary thyroid carcinoma. All published cases in literature were detected incidentally after surgical resection of a cystic mass in the lateral neck. In present case, four lymph nodes were positive for metastases. Papillary carcinoma arising in BCC is able to metastasize to the regional lymph nodes [12], but distant metastasis has not been reported.

In conclusion, the causes of lateral cystic masses in the neck can be complex. They can be congenital, inflammatory, or neoplastic. Nevertheless, the most common lesion is BCC. The clinicians should be cautioned that papillary thyroid

carcinoma arising in this lesion should be taken into consideration. The use of ultrasound and CT parameters is still unable to determine the nature of the lesion, and only pathological examination is required to determine the definitive diagnosis.

TABLE I: SUMMARY OF REPORTED CASES OF PRIMARY THYROID CARCINOMA ARISING IN ECTOPIC THYROID TISSUE WITHIN A BRANCHIAL CLEFT CYST

No	Reference	Age/ Sex	Treatment	Pathologic results for thyroid gland
1	[3]	34/F	Surgical excision of branchial cyst	Thyroidectomy was not done
2	[6]	46/F	Surgical excision of branchial cyst and hemithyroidectomy	Adenomatous goiter, no evidence of carcinoma within the thyroid gland
3	[7]	42/F	Surgical excision of branchial cyst and total thyroidectomy	No evidence of carcinoma within the thyroid gland
4	[8]	36/F	Surgical excision of branchial cyst and total thyroidectomy	Multifocal papillary carcinoma
5	[9]	31/M	Surgical excision of branchial cyst and total thyroidectomy	No evidence of carcinoma within the thyroid gland
6	[10]	29/F	Surgical excision of branchial cyst and total thyroidectomy	No evidence of carcinoma within the thyroid gland
7	[11]	49/M	Surgical excision of branchial cyst, selective right neck dissection, and total thyroidectomy	No evidence of carcinoma within the thyroid gland
8	[12]	41/F	Right lateral neck dissection and total thyroidectomy	Nodular hyperplasia of the right lobe without evidence of carcinoma within the thyroid gland
9	[13]	34/F	Surgical excision of branchial cyst, right modified radial neck dissection, and total thyroidectomy	No evidence of carcinoma within the thyroid gland
10	[14]	35/F	Surgical excision of branchial cyst and total thyroidectomy	No evidence of carcinoma within the thyroid gland
11	[4]	41/F	Surgical excision of branchial cyst; patient refused thyroidectomy	Thyroidectomy was not done
12	[5]	42/M	Surgical excision of the branchial cyst	No thyroidectomy
		29/M	Surgical excision of the branchial cyst	Total thyroidectomy showed no malignancy
		43/M	Surgical excision of the branchial cyst	Total thyroidectomy showed no malignancy
13	Present case, 2021	55/M	Total thyroidectomy and radial neck dissection	Occult follicular variant of thyroid carcinoma

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