

CASE REPORT

Papillary Thyroid Carcinoma within a Second Branchial Cleft Cyst. A Diagnostic and Treatment Challenge

Antonios SKALIAS^a, Konstantinos GAREFIS^{a, b}, Nikolaos TSETOS^a, John GOUDAKOS^a

^aDepartment of Otorhinolaryngology, Head and Neck Surgery,
424 General Military Training Hospital, Thessaloniki, Greece

^b2nd Academic ORL, Head and Neck Surgery Department Aristotle University of Thessaloniki,
Papageorgiou Hospital, Thessaloniki, Greece

**ABSTRACT**

Introduction: Lateral cervical cystic masses are a relatively common occurrence in the otolaryngology practice, which are often attributed to second branchial cleft cysts. However, there should always be a high suspicion for malignancy until proven otherwise.

Case presentation: We present a clinical report of a lateral cervical cyst surgical excision where histopathological examination has revealed features of a branchial cleft cyst harboring a papillary thyroid carcinoma. This led to the clinically latent thyroid primary which was treated through thyroidectomy with selective neck dissection and radioactive iodine therapy.

Discussion: Across current literature regarding thyroid carcinomas arising within branchial cysts, primary thyroid carcinoma was identified in almost half the cases where thyroidectomy was performed. There is no established consensus on the decision of whether to perform a thyroidectomy in such cases, due to the paucity of data for similar cases.

Conclusion: Although branchial cyst is a benign condition, it can harbor malignancy. When thyroid carcinoma occurs within such a cyst, without solid evidence of a thyroid primary, treating physicians are presented with a diagnostic and treatment dilemma. As more cases are discovered and reported, navigating this challenging dilemma will become clearer for the treating physicians.

Keywords: papillary thyroid cancer, papillary thyroid carcinoma, branchial cyst, branchioma, thyroidectomy.

Address for correspondence:

Antonios Skalias, MD, MSc

Department of Otorhinolaryngology - Head and Neck Surgery, 424 General Military Training Hospital, Thessaloniki, Greece

Tel.: +30 2310381738; email: orl.skalias@gmail.com; ORCID: 0000-0001-8517-3071

Article received on the 22nd of April 2024 and accepted for publication on the 17th of September 2024

INTRODUCTION

Second branchial cleft cysts typically present as lateral painless neck masses. Commonly, they are thought to originate from incomplete obliteration of the branchial apparatus during embryogenesis. They are the second most common cause of congenital head and neck mass in children (1). However, neck masses in adults are often caused by malignant neoplasms, therefore warranting caution in their diagnostic approach (2). Cysts or sinuses of the second branchial cleft can exist anywhere along the course of a second branchial cleft sinus, which is as follows: the external opening is located on the neck skin anterior and medial to the sternocleidomastoid, with the fistula traveling deep to platysma then passing in between the internal and external carotids, coursing superficial to both glossopharyngeal and hypoglossal nerves before connecting to the tonsillar fossa.

The first clinical report of papillary carcinoma arising within a branchial cyst was reported in 1992, with 28 similar cases having been reported since then (3-15). Primary thyroid carcinoma was identified in 12 out of the 22 cases in which thyroidectomy was performed. Here we present such a rare clinical occurrence.

CASE PRESENTATION

A 46-year-old patient was referred to our Ear, Nose and Throat Department from a private practice for evaluation and treatment of a mass in the left lateral neck. He had already undergone magnetic resonance angiography (MRA) indicative of a branchial cleft cyst. The mass appeared mobile and tender during the physical examination, with no additional clinical findings and no pertinent medical history. Magnetic resonance angiography revealed a 3 x 2.4 x 3.2 cm well-defined mass located in the left carotid bifurcation, separated by septations, as well as a compact nodule originating on the outer wall of the mass and spreading inwards. The content of the lesion demonstrated a high signal in T1 weighted images, possibly indicative of blood or mucus (Figure 1). There were no other findings from the neck area, concluding that the lesion was possibly a branchial cleft cyst.

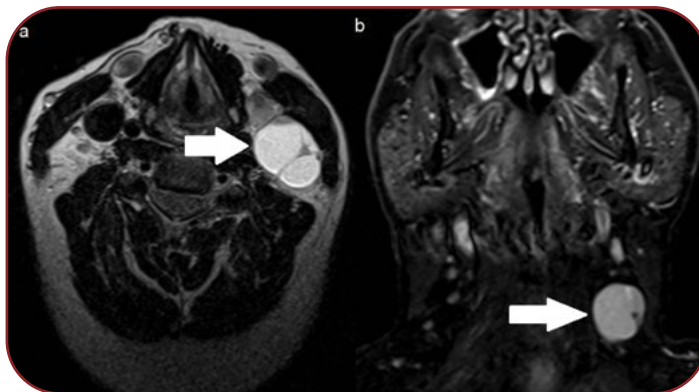


FIGURE 1. Magnetic resonance angiography showing a single mass on the left lateral neck: a) T2 axial; and b) T1 coronal view

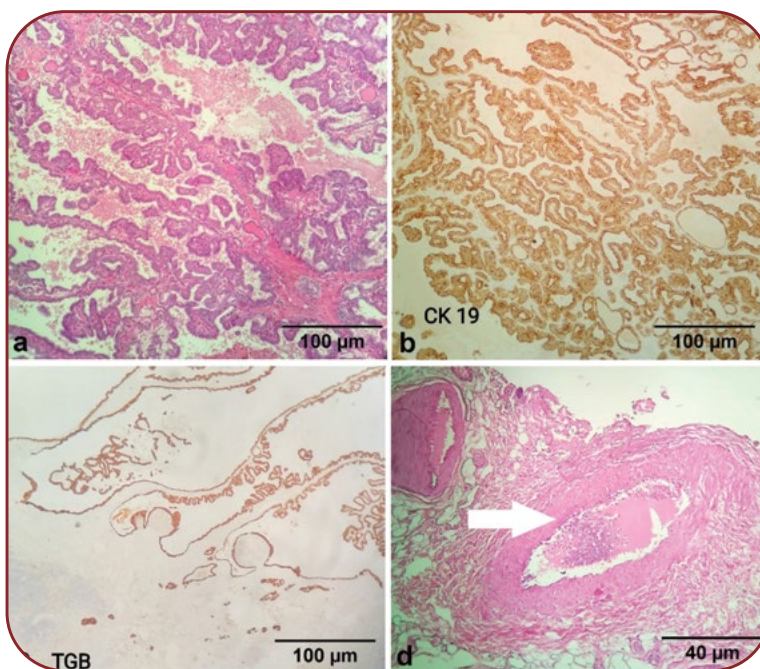


FIGURE 2. Histopathological and immunohistochemical examinations: a) papillary thyroid carcinoma within branchial cleft cyst (H&E stain, x40); b) CK19 immunoreactive; c) TGB immunoreactive; and d) neoplastic emboli within vessel (H&E stain, x100)
H&E=hematoxylin and eosin; CK=cytokeratin; TGB=thyroglobulin

The mass was surgically removed under general anesthesia, without complications, and the excised specimen was forwarded to pathology for further evaluation. No tract was identified traveling from the cyst to the hyoid bone. Histopathological examination revealed histological features of a branchial cleft cyst (Figure 2a-c). Additionally, a papillary carcinoma was observed at a peripheral position, which was immunohistochemically TGB and CK19 positive but CD56 negative.

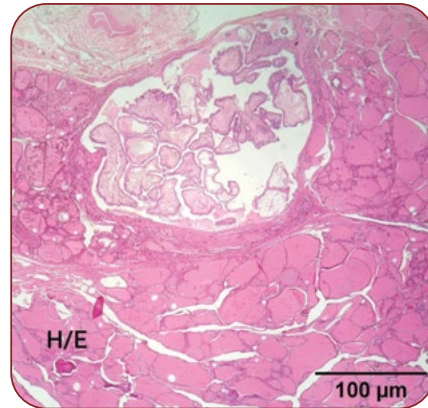


FIGURE 3. Histopathology image of papillary thyroid carcinoma within the thyroid gland (H&E stain, x40). H&E=hematoxylin and eosin

As soon as the pathology results came out, the patient underwent thyroid ultrasound, which revealed a hypoechoic 0.6 x 0.8 cm nodule inferiorly on the isthmus (EU-TIRADS 2), a 0.6 cm cystic nodule in the right lobe (EU-TIRADS 1) and a 0.5 cm cystic nodule in the left lobe (EU-TIRADS 1). Lab results, including calcitonin, FT4, TSH, TG, ANTI-TPO and ANTI-TG, were within normal limits. Subsequent magnetic resonance imaging (MRI) revealed a well-defined 0.5 cm nodular lesion located at the isthmus with benign features. A few lymph nodes at levels II-IV appeared reactively enlarged. Finally, the patient conceded to total thyroidectomy as primary thyroid malignancy could not be ruled out. Total thyroidectomy with selective neck dissection including levels III, IV and VI was performed, under general anesthesia and intraoperative neuro-monitoring, without complications. The postoperative course was uneventful.

Histopathological examination revealed diffuse findings of multinodular goiter with occasional cystic degeneration. A colloid nodule with focal necrosis was reported at the isthmus. A neoplastic population with histological characteristics of papillary thyroid carcinoma, with a maximum diameter of 0.4 cm, was observed on the upper pole of the left lobe, which did not extend beyond the borders of the parenchyma (pT1a) (Figure 3). Two neoplastic emboli were found at uncommon positions within vascular branches (Figure 2d). There was no vascular and capsular invasion or extrathyroidal extension and the surgical margins were clear (the entire thyroid was removed). All lymph nodes had re-

active features. He underwent additional radioactive iodine therapy. There was no recurrence after two years of follow-up.

DISCUSSION

The possibility of thyroid carcinoma arising within lateral neck cysts is rare but not negligible. As a result, several interesting diagnostic and treatment dilemmas are raised. In most of the reported cases, patients initially presented with a lateral cervical mass as the only clinical finding and imaging and/or fine needle aspiration (FNA) did not raise suspicions for malignancy. Is there a method to distinguish cysts harboring malignancy from benign ones? Some authors retrospectively noted imaging features that were possibly indicative of malignancy, such as an isolated and solitary lesion with irregular margins in the lining wall of the cyst, or calcifications (4, 5). In our case, a compact nodule originating on the outer wall of the mass and spreading inwards was noted as well (Figure 1).

After excising the cyst and locating a thyroid carcinoma within, what is the appropriate course of action regarding the thyroid? In most reported cases, there was no conclusive evidence of primary thyroid carcinoma after FNA and/or imaging and/or lab results indicative of abnormal thyroid function. Therefore, treating physicians were presented with the dilemma of whether to perform a thyroidectomy. Sidhu et al suggested that certain features could indicate a carcinoma arising in ectopic thyroid epithelium within the cyst, rather than a thyroid primary (12). In our case, the proposed features were not present (Figure 2) and thyroid examination revealed a thyroid primary indeed. Many more cases need to be reported and their findings synthesized in order to reach a consensus on when to perform a thyroidectomy.

Twenty-eight cases of papillary carcinoma arising within a branchial cyst have been reported in the current literature (3-15). In 22 cases, thyroidectomy was performed. Primary thyroid carcinoma was identified in 12 out of those 22 cases. Selective neck dissection was reported in most of the cases.

Additionally, the analysis of the presence of thyroid carcinoma in a lateral neck cyst with regard to the existence of a primary thyroid tumor or lack thereof could contribute to the

discussion of lateral neck cysts' embryologic origin (11). Reporting the presence of an associated tract or lack thereof in future clinical reports could be important as well (9, 12).

CONCLUSIONS

Cervical masses in adults are a common occurrence. Their causes are often benign; however, there should always be a high suspicion of malignancy, until proven otherwise. Although a branchial cyst is a benign condition, it can harbor malignancy. When thyroid carcinoma occurs within such a cyst, without solid evidence of a thyroid primary, treating physicians are pre-

sented with a diagnostic dilemma. Currently, there is no established consensus for the further treatment of such cases due to the paucity of data for similar cases. In our case, thyroidectomy revealed a primary papillary thyroid carcinoma; however, this is not always the case. As more cases are discovered and reported, metanalyses may serve to uncover more details regarding the incidence of thyroid carcinomas within lateral neck cysts, and the features that indicate a possible relationship between those and primary thyroid malignancy. □

Conflicts of interest: none declared.

Financial support: none declared.

REFERENCES

1. Goff CJ, Allred C, Glade RS. Current management of congenital branchial cleft cysts, sinuses, and fistulae. *Curr Opin Otolaryngol Head Neck Surg* 2012;20:533-539.
2. Pynnonen MA, Gillespie MB, Roman B, et al. Clinical Practice Guideline: Evaluation of the Neck Mass in Adults. *Otolaryngol Head Neck Surg* 2017;157:S1-S30.
3. Marotta DA, Morley TJ, Jabaay MJ, et al. Branchial Cleft Cyst Harbors Metastatic Papillary Thyroid Carcinoma. *Cureus* 2021;13:e13940.
4. Cooc A, Chong I, Wang KY, et al. Papillary thyroid carcinoma metastasis to a branchial cleft cyst: a case report and review of imaging. *Clin Imaging* 2020;64:1-6.
5. Hong YT, Hong KH. Radiologic Features of Papillary Carcinoma in the Second Branchial Cleft Cyst. *Ear Nose Throat J* 2019;98:295-298.
6. Gür H, Arpacı RB, İsmi O, et al. Papillary Thyroid Carcinoma Spreading into Branchial Cleft Cyst. *Turk Arch Otorhinolaryngol* 2019;57:95-98.
7. Tazegul G, Bozoğlan H, Doğan Ö, et al. Cystic lateral neck mass: Thyroid carcinoma metastasis to branchial cleft cyst. *J Cancer Res Ther* 2018;14:1437-438.
8. Papadakis CE, Ladias A, Chimona TS, et al. Thyroid Papillary Carcinoma in a Branchial Cleft Cyst—A Case Report. *Journal of Cancer Therapy* 2017;08:278-285.
9. Xu JJ, Kwan K, Fung K. Papillary thyroid carcinoma in a lateral neck cyst: primary of ectopic thyroid tissue versus cystic metastasis. *J Laryngol Otol* 2013;127:724-727.
10. Sagit M, Gokler A, Akin I, Han U. Wrong egg in the usual nest: thyroid papillary carcinoma within a branchial cleft cyst. *Ear Nose Throat J* 2013;92:E31-E34.
11. Ruhl DS, Sheridan MF, Sniezek JC. Papillary Thyroid Carcinoma in a Branchial Cleft Cyst without a Thyroid Primary: Navigating a Diagnostic Dilemma. *Case Rep Otolaryngol* 2013;2013:405342.
12. Sidhu S, Lioe TF, Clements B. Thyroid papillary carcinoma in lateral neck cyst: missed primary tumour or ectopic thyroid carcinoma within a branchial cyst? *J Laryngol Otol* 2000;114:716-718.
13. Yehuda M, Schechter ME, Abu-Ghanem N, et al. The incidence of malignancy in clinically benign cystic lesions of the lateral neck: our experience and proposed diagnostic algorithm. *European Archives of Oto-Rhino-Laryngology* 2018;275:767-773.
14. Seven H, Gurkan A, Cinar U, et al. Incidence of occult thyroid carcinoma metastases in lateral cervical cysts. *Am J Otolaryngol* 2004;25:11-17.
15. Rigas G, Skalias A, Filentas A, et al. An Unusual Case of a Papillary Thyroid Carcinoma Arising in a Branchial Cyst. *Ear Nose Throat J* 2023;1455613231207819..