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Upper Neck Papillary Thyroid Cancer: A New Proposed Term for the Composite of Thyroglossal Duct Cyst-Associated Papillary Thyroid Cancer, Pyramidal Lobe Papillary Thyroid Cancer, and Delphian Node Papillary Thyroid Cancer Metastasis

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Objectives/Hypothesis: Thyroglossal duct cyst (TGDC) is a common congenital anomaly, but TGDC carcinoma is rare. Thyroglossal duct cyst carcinoma management is controversial, especially that of the orthotopic thyroid gland. We aim to provide an insight into the pathologic basis of this management controversy through the review of 28 TGDC cancer cases, thus far the largest such series to our knowledge.

Study Design: Retrospective.

Methods: Twenty-eight cases recorded as TGDC cancer in the hospital database were reviewed; their initial clinical diagnosis from medical chart review (DX1) and final pathological review diagnosis (DX2) through pathology slides review by our pathologist (blinded to DX1) were compared. The thyroid gland management and pathology were evaluated.

Results: In the 28 TGDC carcinoma (hospital-recorded diagnosis) patients, DX1 and DX2 were respectively reported as 53% and 14% TGDC carcinoma, 11% and 29% as pyramidal lobe primary, and 4% and 25% as metastatic Delphian node. Thirty-two percent of cases were in the indeterminate category, in both DX1 and DX2, but included different patients. Thyroidectomy was performed in 54% of the cases, papillary thyroid cancer (PTC) was reported in 37% of these thyroid glands. Concurrent thyroid gland malignancy was reported in all Delphian node and pyramidal lobe PTC patients.

Conclusion: The diagnosis of TGDC cancer comprises a heterogeneous group that includes true TGDC cancer, pyramidal lobe primary, Delphian node metastasis, and indeterminate cases. We propose a new terminology of upper neck papillary thyroid carcinoma (UPTC) to denote this heterogeneous group and recommend a rational algorithm for management. Correct pathologic subcategory and thyroid ultrasonography are essential for optimal management of thyroid gland in UPTC cases.

Key Words: Thyroglossal duct cyst cancer, pyramidal lobe PTC, Delphian node metastasis, midline neck mass.

Level of Evidence: 4.

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INTRODUCTION

Thyroglossal duct cyst (TGDC), a common congenital anomaly related to thyroid gland development, presents as a midline neck mass.[1] Although TGDC is common, primary carcinoma of the thyroglossal duct cyst (TGDC cancer) is rare, reported in less than 1% of TGDC patients. Thyroglossal duct cyst carcinoma is most typically papillary thyroid cancer (PTC).[2] The first case of TGDC carcinoma was reported by Brentano in 1911.[3] Since then, approximately 250 cases have been reported, the vast majority as single case reports or small case series.[4] Currently, there is no consensus on optimal management of TGDC cancer. Controversy mostly centers on the role of thyroidectomy, as well as lymph node dissection, postoperative thyroid suppression, and radioiodine treatment.

Even with careful clinical and histologic review, we often have found it difficult to distinguish true TGDC cancer from other similar but discrete diagnoses, including pyramidal lobe PTC and Delphian node (i.e. prelaryngeal lymph node) PTC metastasis. This is not surprising, given the significant clinical and radiological similarities associated with these three diagnoses—all of these entities represent foci of PTC, typically cystic and presenting as midline upper neck nodular lesions. The difficulty in distinguishing these entities may provide an explanation for the observed heterogeneity in reports of TGDC cancers and for some of the controversies in its management, especially as it relates to the need for thyroidectomy.

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TABLE I.
Key Histology Features Considered for Obtaining Final Pathologic Diagnosis (DX2) of TGDC Cancer, Pyramidal Lobe Cancer, and Delphian Node Metastasis.

DX2 Final Pathologic Diagnosis	Key Features of Primary Indexed Midline Upper Neck Lesion
TGDC cancer	The presence of benign respiratory, cuboidal, or squamous epithelial cyst lining in association with the cystic PTC, and
	Lack of features of lymph node architecture, and
	Lack of evidence of a primary PTC within thyroid parenchyma
Delphian node metastasis	Histologic features of PTC with lymph node architecture, including lymphoid stroma and subcapsular sinus
Pyramidal lobe primary	PTC surrounded by a background of benign thyroid parenchyma, and
	Lack of lymph node architecture or TGDC features described above
Indeterminate origin	Exact diagnosis based on above key features could not be achieved.

DX2 = final pathological review diagnosis; LN = lymph node; PTC = papillary thyroid cancer; TGDC = thyroglossal duct cyst.

We present a series of 28 patients with a hospital-recorded diagnosis of TGDC cancer, to our knowledge the largest series of such patients in the literature. In-depth clinical and pathological review by the senior author (G.W.R.) and re-evaluation of the pathology slides by our thyroid pathologist (W.F.) were performed. This review revealed that these cases were in fact a heterogeneous group of entities. We therefore propose a new terminology of upper neck papillary cancer (UPTC) representing these heterogeneous entities, namely: 1) TGDC cancer; 2) pyramidal lobe cancer; 3) Delphian node metastasis, and 4) indeterminate origin (no definitive diagnosis achieved). Thus, UPTC incorporates all of the pertinent diagnoses appropriate for a clinician to consider in managing this heterogeneous group, which previously have been diagnosed as TGDC cancer. We provide an algorithm for the management of UPTC after initial surgery.

MATERIALS AND METHODS

After obtaining institutional review board approval, 575 TGDC patients were identified by searching hospital pathology data bases from 1992 to 2013 at the Massachusetts Eye and Ear Infirmary and the Massachusetts General Hospital, Boston, Massachusetts . From these 575 patients, benign TGDC cases were excluded to yield 28 cases of TGDC cancer (hospital-recorded diagnosis). A detailed retrospective review including patient demographics, history and physical exam, imaging results, treatment, pathological features, and outcome data from the medical records of these 28 patients was performed by our senior clinician-surgeon (G.W.R.) to generate the initial clinical diagnosis (DX1). Re-evaluation of surgical pathology slides by a single thyroid pathologist from our institution (W.F.), who was blinded to DX1, generated the final pathological review diagnosis (DX2). This pathologist is the chief of head, neck, and thyroid pathology at our tertiary care institution. The DX1 was compared to the DX2. The DX2 was based on the finding of multiple histologic features (shown in Table I), interpreted within the overall context of the case.

Kappa coefficient to evaluate agreement between DX1 and DX2 was calculated using SAS 9.4 (SAS Institute Inc., Cary, NC)

RESULTS

Demographics, Clinical Presentation, and Preoperative Evaluation

After review of pathology hospital data bases, 575 TGDC cases were identified and 28 patients (4.9%) with TGDC cancer were included in the study. The mean age of these patients was 40 years (range 22–61 years). There were 17 females and 11 males, with a male-to-female ratio of 1:1.6.

Asymptomatic neck mass was the most common presentation (27 of 28); movement with deglutition was noted in eight patients; six patients had an abnormal thyroid exam; and eight patients had positive family history of benign thyroid disease. Other less common symptoms were dysphagia and prior infection of the index lesion. Dyspnea, history of fistula, and palpable neck nodes were not present in any of the patients. Preoperative labs including T3, T4, and TSH were normal in all cases. Preoperative imaging studies are summarized in Table II. Out of nine patients in whom fine needle aspiration (FNA) biopsy was performed, two were malignant (PTC), three were benign, and four were suspicious for malignancy or showed atypical cells.

Surgical Treatment

In patients undergoing surgical treatment for the first time (23 patients), the Sistrunk procedure was performed in 70%, simple cyst excision in 12%, and total thyroidectomy with nodal dissection in 18%. Among revision surgery patients (5 patients), total thyroidectomy with nodal dissection was performed in three; Sistrunk procedure with nodal dissection and only Sistrunk procedure were performed in one each.

Surgical Pathology

Mean size of UPTC lesions was 1.94 cm. (range 0.1–4.5cm). Histology was reported as classical PTC in 50% of cases (14/28), classical cystic in 46.4% (13/28) (one with squamous metaplasia), and follicular variant of

TABLE II.
Preoperative Imaging Results.

Preoperative Radiological Exams:	Significant Findings
Ultrasound, N = 10	Thyroid abnormalities (nodules) noted in 5 cases
Computed tomography scan, N = 12	Cystic index lesion noted in 6 cases
Magnetic resonance imaging, N = 2	Thyroid nodule noted in 1 case
	Hemorrhagic cyst suspicious of malignancy noted in 2nd case
Radionuclide scan, N = 1	Normal thyroid gland

TABLE III.
Lymph Node Location and Positivity in Our Series.

Lymph Nodes Positivity	35.7%
Mean Number and Range	Mean = 1.5 lymph nodes
	Range (1–4 lymph nodes)
Positive Lymph Node Location	Central: 91%
	Lateral: 9%

PTC in 3.6% (1/28). Well-developed nuclear features of PTC were noted in all cases. In patients with TGDC PTC, benign TGDC epithelium that was predominantly ciliated and columnar was present. Invasion of TGDC wall was observed in 67% of cases and lymphocytes were present in 37%.

When thyroid surgery was performed, 42% of cases had a normal thyroid gland; 37% contained PTC; 16% had Hashimoto's thyroiditis; and 5% had benign adenoma. Twenty-one patients underwent lymph node dissection either at the time of initial surgery or subsequently. The lymph node location and presence of metastatic disease are denoted in Table III.

Initial Clinical and Surgical Pathology Reviews (DX1) Versus Pathology Re-Review (DX2)

All patients had a recorded diagnosis of TGDC cancer based on hospital database entry. Initial clinical diagnosis (DX1) obtained by clinical review resulted in the following diagnostic assessment: TGDC carcinoma in 53% of patients (15/28), pyramidal lobe primary in 11% (3/28), metastatic midline Delphian lymph node in 4% (1/28), and indeterminate (no definitive clinical diagnosis) in 32% (9/28).

Final pathological review diagnosis was obtained by reevaluation of the pathology slides by a pathologist with a special interest and expertise in thyroid pathology. It yielded the following pathologies: TGDC carcinoma in 14% of patients (4/28), pyramidal primary in 29% (8/28), metastatic midline Delphian lymph node in 25% (7/28), and indeterminate origin in 32% (9/28) (Fig. 1). Table IV shows the distribution of initial clinical diagnosis (DX1) for each subcategory of UPTC compared to the final pathological review diagnosis (DX2). The simple Kappa coefficient (*K* value) to check agreement between DX1 and DX2 was found to be 0.1765 (Table IV), demonstrating poor agreement between DX1 and DX2.[5]

Follow-Up

The mean follow-up period was 56 months (range 1–180 months). Eleven patients had subsequent total thyroidectomy; of those, Sistrunk procedure was performed in 10 patients and cyst excision in one patient. When all thyroid surgeries (initial and subsequent) were grouped together, a total of 17 thyroidectomies were performed, of which six cases (37%) were positive for PTC overall. The distribution of thyroid surgery in each subcategory of UPTC based on DX2 and the presence of thyroid malignancy are shown in Table V.

No patient had local, regional, or distant recurrence. Radioactive iodine (RAI) was administered in 12 patients with a dose ranging between 30 and 100 mCi. Thyroid suppression was achieved in all thyroid surgery patients.

DISCUSSION

Although TGDC is one of the most frequent congenital anomalies of the neck, the incidence of TGDC cancer is low. Current series differ in treatment and postoperative follow-up recommendations. Our findings of female predominance and asymptomatic neck mass as the most common presenting symptom support results of the other studies. Although the rate of TGDC cancer in the literature is reported as 1% to 2%, higher rates have also been reported; our rate of 4.9% may represent the bias of a tertiary care center.[6,7]

UPTC and the Diagnostic Heterogeneity of the Initial TGDC Cancer Diagnosis

In our series, a detailed pathologic review was important to ultimately render a specific diagnosis that could lead to focused optimal treatment. Although other studies discuss the importance of the proper diagnosis, none report the diagnostic variability in collected data of apparent TGDC cancer when the cases are subject to additional intense clinical and pathologic review.[8]

Our series represents a large cohort with an initial hospital-recorded diagnosis of TGDC cancer. Through

TABLE IV.
Distribution of UPTC Subcategories of Initial Clinical Diagnosis Within Final Pathological Review Diagnosis and Kappa Coefficient Value.

	Final Pathological Review Diagnosis (pathology slides review-DX 2)			
Initial Clinical Diagnosis (DX 1)	TGDC Cancer 4 cases (14%)	Pyramidal Primary 8 cases (29%)	Metastatic Midline LN 7 cases (25%)	Indeterminate Origin 9 cases (32%)
TGDC cancer 15 cases (53%)	4	1	4	6
Pyramidal primary 3 cases (11%)	0	2	1	0
Metastatic midline LN 1 case (4%)	0	0	1	0
Indeterminate origin 9 cases (32%)	0	5	1	3

Simple Kappa coefficient (*K* value) for DX1 and DX2 agreement = 0.1765, 95%; confidence limits = −0.0185 to 0.3714.
DX1 = initial clinical diagnosi; DX2 = final pathological review diagnosis; LN = lymph node; TGDC = thyroglossal duct cyst.

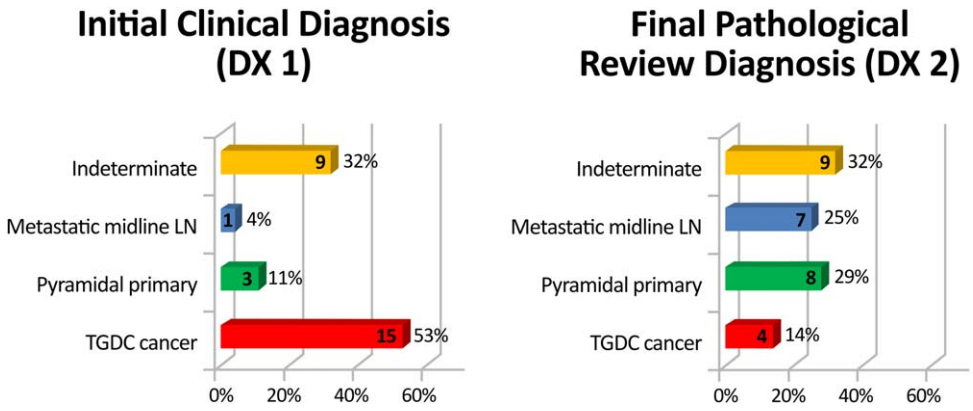

Fig. 1. Graph showing percentages of different subcategories of UPTC in initial clinical diagnosis (DX 1) and in final pathological review diagnosis (DX 2).
DX1 = initial clinical diagnosis; DX2 = final pathological review diagnosis; TGDC = thyroglossal duct cyst; UPTC = upper neck papillary thyroid carcinoma. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

detailed clinical and pathologic review, these patients actually represented three distinct diagnoses: TGDC cancer, pyramidal lobe PTC, and Delphian node metastasis—as well as a fourth indeterminate category. It should be noted that sometimes thyroid cancer metastasis in a node may degrade the node, and the nodal architecture may be unapparent to the pathologist. This may be the dynamic in some of our cases in the indeterminate category.

The interpretation of all existing series of TGDC cancer should therefore suspect that these series likely contain the same diagnostic heterogeneity as our patients. The fact that careful clinical and pathologic review still resulted in an indeterminate diagnosis in 32% cases underscores the diagnostic challenge that these patients represent. Given this diagnostic challenge, we propose that these patients should be grouped together preoperatively as upper (midline) neck PTC or UPTC. The term *UPTC* emphasizes the various diagnoses needing consideration in such patients as they are evaluated and treated, as well as provides an overarching diagnostic framework for management.

Pathological review of the UPTC surgical specimen should be exhaustive to allow subcategorization into TGDC cancer, pyramidal lobe cancer, Delphian node metastasis, or indeterminate. Our pathologic re-evaluation (DX2) revealed a surprisingly different subcategorization of the UPTC group. The Kappa coefficient showed poor agreement between clinical DX1 and final pathologic DX2. Almost 40% of patients who were initially labeled as TGDC-associated PTC were in fact ultimately diagnosed as Delphian node metastases or pyramidal lobe primaries. From the DX1 to the DX2, the rate of pyramidal lobe primaries upsurged from 11% to 29%, and the rate of Delphian node metastases upsurged from 4% to 25%.

The Exact UPTC Subcategory Diagnosis and Need for Thyroid Surgery

The clinical significance of the exact subcategory diagnosis of UPTC group has great importance in deciding the need for surgical excision of the orthotopic thyroid gland. In our series, concurrent PTC in the thyroid gland was not found in any of the four true TGDC cancer patients who underwent thyroid surgery. Although one would presume that true TGDC cancer is infrequently associated with orthotopic thyroid gland PTC, the rate of orthotopic thyroid gland PTC for larger series of TGDC cancer in the literature is approximately 1/3. This fits very well with our data for our entire cohort reporting thyroid gland PTC in 37% of cases. Hartl et al., in a series of 18 TGDC cancers, reported additional tumor foci in the thyroid gland in more than half of the patients.[9] The Mayo Clinic, in a study of 12 TGDC cancers, reported thyroid involvement in 33% of cases.[10] Other series have reported 20% to 62% of incidences of thyroid involvement.[2,8,11,12] Our findings suggest that, in these series, some of the reported TGDC cancers are likely pyramidal lobe PTC or Delphian nodes metastasis.

The need for orthotopic thyroid gland excision thus depends on the UPTC subcategory determination. In our opinion, it should also take into account the orthotopic thyroid ultrasound (US) findings. Concurrent PTC in the thyroid gland was not found in any of the four true TGDC cancer patients in our series who underwent thyroid surgery. Hence, when the thyroid US is negative and the diagnosis is clearly TGDC cancer, thyroidectomy can be avoided (assuming RAI is not planned based on

TABLE V.
Number of Initial/Subsequent Thyroid Surgeries Performed in Each Subcategory of UPTC (based on DX 2) and Presence of Thyroid Malignancy.

Final Pathological Review Diagnosis (DX 2)	No. of Cases With Thyroid Surgery	Thyroid Malignancy (PTC)
TGDC cancer (N = 4)	4	0
Pyramidal (N = 8)	2	1 (50%)
Metastatic delphian midline LN (N = 7)	4	3 (75%)
Indeterminate origin (N = 9)	7	1 (16%)

DX2 = final pathological review diagnosis; LN = lymph node; PTC = papillary thyroid cancer; TGDC = thyroglossal duct cyst; UPTC = upper neck papillary thyroid carcinoma.

the histology of the TGDC cancer itself) (Fig. 2). However, in Delphian node metastases patients, all but one patient had orthotopic gland PTC in our series. We suspect that in the remaining case the thyroid gland primary may be occult or regressed. Thyroidectomy is also recommended in cases of a pyramidal lobe primary (Fig. 2). Reports of PTC multifocality range from 24% to 80%.[13–16] In our series, 16% of cases in whom the cancer was of indeterminate origin had orthotopic thyroid gland cancer. Hence, these cases need to be managed on an individual basis. We elect to suggest consideration of thyroidectomy in this group (Fig. 2)

We acknowledge that the preoperative diagnosis of cancer in TGDC patients is difficult. However, with proper pathologic review of the surgical specimen, an exact subcategory diagnosis of UPTC cases into TGDC cancer, pyramidal lobe cancer, or Delphian node metastasis can be obtained in a majority of cases. Accurate UPTC subcategory diagnosis facilitates the decision making regarding thyroidectomy and should incorporate the orthotopic thyroid gland US results. Thyroid surgery therefore should be considered in all patients who are diagnosed with Delphian node PTC or pyramidal lobe PTC, and could be considered in those patients with UPTC of indeterminate origin. In patients who have a final diagnosis confirmed as TGDC-associated PTC, one may safely avoid thyroid surgery if:

1. The primary characteristics of the TGDC-associated PTC as it relates to size, degree of invasion, and nodal status would not require RAI. If RAI is judged necessary, then thyroidectomy would be required.[10,17,18]
2. Those patients with TGDC-associated PTC, who do not require RAI, may still require thyroid surgery if there is significant thyroid gland nodularity on US. One could perform empiric thyroidectomy or consider US-guided FNA assessment. If FNA is negative and the patient undoubtedly has TGDC-associated PTC that would not require RAI, then we believe that the patient can be safely followed without thyroidectomy. Other researchers agree that US-negative patients can be followed without thyroid surgery.[6,10,11]

Preoperative Evaluation

Preoperative diagnosis of carcinoma in a patient presenting with TGDC is unusual; in most cases, the diagnosis of TGDC cancer is not made until after surgery. Indeed, carcinoma is usually indistinguishable from benign TGDC in terms of localization, size, and other physical exam characteristics. We were rarely able to establish a diagnosis of carcinoma preoperatively, with only 7% patients being preoperatively diagnosed as carcinoma. Diagnostic sensitivity of FNA was only 22.2%, probably due to the low sensitivity of FNA for cystic lesions. We were unable to raise the suspicion of malignancy based on US and computed tomography (CT) scan unless lymph node disease was identified. Ultrasonography characteristics that suggest TGDC cancer include the presence of a mural mass with associated microcalcifications.[19] Computed tomography and magnetic resonance imaging images suggestive of carcinoma are the presence of a dense or enhancing mural nodule,

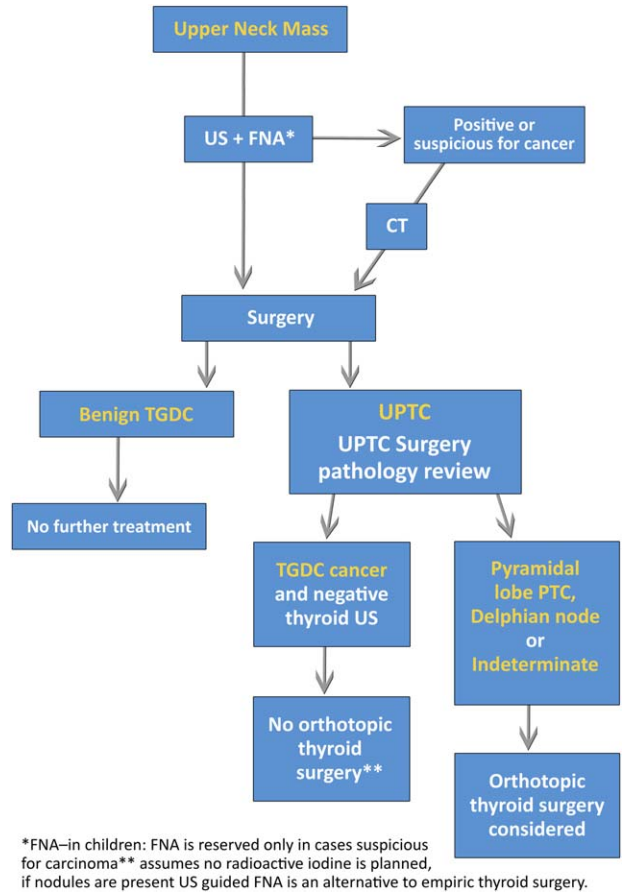

Fig. 2. Algorithm for preoperative evaluation and management of a patient with upper neck mass.
CT = computed tomography; FNA = fine needle aspiration; PTC = papillary thyroid cancer; TGDC = thyroglossal duct cyst; UPTC = upper neck papillary thyroid carcinoma; US = ultrasound. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

isolated calcification, and an irregular margin.[20–22] Results of TGDC malignancy obtained on FNA vary in the literature. Pellegriti et al. reported the preoperative diagnosis of TGDC carcinoma in only seven out of 26 patients (27%); cytological suspicion of malignancy based on FNA was established in an additional five patients.[23] Generally, FNA is reported as a diagnostic tool with a low diagnostic accuracy due to the frequent cystic nature of the lesions.[8,24] Some studies support FNA for TGDC only in certain cases because of the low frequency of occurrence of carcinoma.[25] In some studies, however, FNA was performed in all TGDC cases, and authors have reported much higher sensitivity and specificity.[8,10,26,27]

We propose an algorithm for the preoperative evaluation of patients with TGDC, which represents a modified algorithm of Pribitkin and Friedman.[28] In patients with an upper neck mass noted on physical exam, we propose at minimum a neck US. A CT scan is also often useful in the evaluation of the mass and its relationship to surrounding structures. Ultrasound should be coupled with FNA in adult patients; among children, FNA is limited to cases when there is a high suspicion of malignancy based on US and CT characteristics (Fig. 2). Ultrasound and CT

should also evaluate the status of the thyroid gland, central neck, and lateral neck nodal basins.[29]

CONCLUSION

Upper neck papillary thyroid carcinoma represents a clinical diagnosis that consists of a heterogeneous group composed of pyramidal lobe primary, Delphian node metastases, and TGDC carcinoma. Our series suggests that much of what is known and previously published about TDGC cancer is in fact a representation of several distinct entities with varying clinicopathologic behaviors. Much of the controversy in managing the thyroid gland in these patients in the literature may be explained by this diagnostic heterogeneity. The correct pathologic subcategory diagnosis after initial surgery in a UPTC patient allows for optimal management, especially as it relates to the recommendation for thyroid surgery.

LIMITATIONS

Although our pathologist has expertise that may not be readily available in all centers, the key histology criteria considered for obtaining final pathological diagnosis (Table I) can assist pathologists in categorizing the index lesion and should be helpful in minimizing individual variations that may occur.

BIBLIOGRAPHY

1. Shindo M. Ectopic thyroid and thyroglossal duct cysts. In: Randolph G, ed. Surgery of the Thyroid and Parathyroid Glands. 2nd ed. Philadelphia, PA: Elsevier; 2013: 60–62.
2. Weiss SD, Orlich CC. Primary papillary carcinoma of a thyroglossal duct cyst: report of a case and literature review. *Br J Surg* 1991;78:87–89.
3. Brentano H. Struma aberrata lingual mit druzen metastasen. *Dtsch Med Wochenschr* 1911;37:665–666.
4. Dzodic R, Markovic I, Stanojevic B, et al. Surgical management of primary thyroid carcinoma arising in thyroglossal duct cyst: an experience of a single institution in Serbia. *Endocr J* 2012;59:517–522.
5. Altman DG. Practical Statistics for Medical Research. London, UK: Chapman and Hall; 1991.
6. Patel SG, Escrig M, Shaha AR, et al. Management of well-differentiated thyroid carcinoma presenting within a thyroglossal duct cyst. *J Surg Oncol* 2002;79:134–139; discussion 140–131.
7. Forest VI, Murali R, Clark JR. Thyroglossal duct cyst carcinoma: case series. *J Otolaryngol Head Neck Surg* 2011;40:151–156.
8. Plaza CP, Lopez ME, Carrasco CE, Meseguer LM, Perucho Ade L. Management of well-differentiated thyroglossal remnant thyroid carcinoma: time to close the debate? Report of five new cases and proposal of a definitive algorithm for treatment. *Ann Surg Oncol* 2006;13:745–752.
9. Hartl DM, Al Ghuzlan A, Chami L, Leboulleux S, Schlumberger M, Travagli JP. High rate of multifocality and occult lymph node metastases in papillary thyroid carcinoma arising in thyroglossal duct cysts. *Ann Surg Oncol* 2009;16:2595–2601.
10. Heshmati HM, Fatourechi V, van Heerden JA, Hay ID, Goellner JR. Thyroglossal duct carcinoma: report of 12 cases. *Mayo Clin Proc* 1997;72: 315–319.
11. Doshi SV, Cruz RM, Hilsinger RL Jr. Thyroglossal duct carcinoma: a large case series. *Ann Otol Rhinol Laryngol* 2001;110:734–738.
12. Miccoli P, Minuto MN, Galleri D, Puccini M, Berti P. Extent of surgery in thyroglossal duct carcinoma: reflections on a series of eighteen cases. *Thyroid* 2004;14:121–123.
13. Shah JP, Loree TR, Dharker D, Strong EW, Begg C, Vlamis V. Prognostic factors in differentiated carcinoma of the thyroid gland. *Am J Surg* 1992;164:658–661.
14. Hay ID, Grant CS, Bergstralh EJ, Thompson GB, van Heerden JA, Goellner JR. Unilateral total lobectomy: is it sufficient surgical treatment for patients with AMES low-risk papillary thyroid carcinoma? *Surgery* 1998;124:958–964; discussion 964–956.
15. Machens A, Holzhausen HJ, Dralle H. The prognostic value of primary tumor size in papillary and follicular thyroid carcinoma. *Cancer* 2005; 103:2269–2273.
16. Witt RL. Initial surgical management of thyroid cancer. *Surg Oncol Clin N Am*. 17:71–91, viii.
17. Kennedy TL, Whitaker M, Wadih G. Thyroglossal duct carcinoma: a rational approach to management. *Laryngoscope* 1998;108:1154–1158.
18. Miccoli P, Minuto MN, Galleri D, Puccini M, Berti P. Extent of surgery in thyroglossal duct carcinoma: reflections on a series of eighteen cases. *Thyroid* 2004;14:121–123.
19. Cignarelli M, Ambrosi A, Marino A, Lamacchia O, Cincione R, Neri V. Three cases of papillary carcinoma and three of adenoma in thyroglossal duct cysts: clinical-diagnostic comparison with benign thyroglossal duct cysts. *J Endocrinol Invest* 2002;25:947–954.
20. Branstetter BF, Weissman JL, Kennedy TL, Whitaker M. The CT appearance of thyroglossal duct carcinoma. *AJNR Am J Neuroradiol* 2000;21: 1547–1550.
21. Samara C, Bechrakis I, Kavadias S, Papadopoulos A, Maniatis V, Strigaris K. Thyroglossal duct cyst carcinoma: case report and review of the literature, with emphasis on CT findings. *Neuroradiology* 2001;43:647–649.
22. Glastonbury CM, Davidson HC, Haller JR, Harnsberger HR. The CT and MR imaging features of carcinoma arising in thyroglossal duct remnants. *AJNR Am J Neuroradiol* 2000;21:770–774.
23. Pellegriti G, Lumera G, Malandrino P, et al. Thyroid cancer in thyroglossal duct cysts requires a specific approach due to its unpredictable extension. *J Clin Endocrinol Metab* 2013;98:458–465.
24. Shahin A, Burroughs FH, Kirby JP, et al. Thyroglossal duct cyst: a cytopathologic study of 26 cases. *Diagn Cytopathol* 2005;33:365–369.
25. Auguste LJ, Attie JN. Completion thyroidectomy for initially misdiagnosed thyroid cancer. *Otolaryngol Clin North Am* 1990;23:429–439.
26. Miccoli P, Minuto MN, Ugolini C, et al. Intrathyroidal differentiated thyroid carcinoma: tumor size-based surgical concepts. *World J Surg* 2007; 31:888–894.
27. Luna-Ortiz K, Hurtado-Lopez LM, Valderrama-Landaeta JL, Ruiz-Vega A. Thyroglossal duct cyst with papillary carcinoma: what must be done? *Thyroid* 2004;14:363–366.
28. Pribitkin EA, Friedman O. Papillary carcinoma in a thyroglossal duct remnant. *Arch Otolaryngol Head Neck Surg* 2002;128:461–462; discussion 463–464.
29. Lesnik D, Cunnane ME, Zurakowski D, et al. Papillary thyroid carcinoma nodal surgery directed by a preoperative radiographic map utilizing CT scan and ultrasound in all primary and reoperative patients. *Head Neck* 2014;36:191–202.