Thyroglossal Duct Cyst Carcinomas in Pediatric Patients: Report of Two Cases with a Comprehensive Literature Review

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Abstract Thyroglossal duct cyst (TGDC) carcinomas are rarely encountered in the pediatric population. The clinical behavior of these tumors in the pediatric setting is unclear and management is not well defined. Two cases of pediatric thyroglossal duct cyst carcinoma were identified in a review of all thyroglossal duct cysts diagnosed over a ten year period. These two cases were analyzed along with 57 cases of thyroglossal duct cyst carcinoma affecting patients less than 21 years of age compiled from the English literature. Fifty-nine patients (36 females, 23 males) aged 6–20 years (mean 15.0 years) were identified. All presented with an anterior midline neck mass, which was typically mobile and non-tender. The average tumor size was 2.6 cm. Histologically, all tumors were papillary thyroid carcinomas arising in a background of a thyroglossal duct cyst. The tumors exhibited a papillary, follicular, or mixed architecture with classical papillary thyroid carcinoma nuclear features. Soft tissue extension was present in 16 cases. All patients were managed surgically with a Sistrunk procedure, with additional thyroidectomy performed in 29 patients, combined with a lymph node dissection (n=15), or a Sistrunk and lymph node dissection (n=5). All patients were stage I at presentation, with 11 showing lymph node metastases. Postoperative radioactive iodine was employed in 20 patients. A papillary carcinoma of the thyroid gland was reported in four of the patients

who had concurrent/subsequent thyroidectomies. Recurrences were reported in four patients, with distant metastases in one patient, who died of disease (13 months). Follow up data was available for 45 patients, with an overall mean follow-up of 54.5 months. All patients were alive, with the exception of one who died with disease. TGDC carcinomas in pediatric patients is associated with a good overall prognosis, best managed by Sistrunk procedure alone, with selected lymph node dissection if clinically indicated.

Keywords Carcinoma, papillary · Thyroglossal duct cyst · Incidence · Follow-up studies · Prognosis · Thyroid neoplasms · Thyroidectomy · Pediatric · Childhood · Adolescence

Introduction

Thyroglossal duct cysts (TGDCs) are well recognized lesions arising from remnants of the thyroglossal duct, which forms during embryogenesis of the thyroid gland. They are commonly encountered in the midline neck region and may manifest at any age [1–3]. Rarely, TGDCs may harbor an associated carcinoma. The incidence of malignancy among TGDCs is difficult to define, though a recent large community practice based series suggests a frequency of approximately 3% [4]. TGDC carcinomas occur predominantly in adults, typically in the fourth decade of life [1, 4]. Although thyroglossal duct cysts frequently affect children and adolescents, thyroglossal duct cyst carcinomas are rare in the pediatric age group, with less than 60 cases reported to date [4–53].

Due to the rare occurrence of TGDC carcinoma in children and adolescents, the biologic behavior of this malignancy in the pediatric age group remains unclear and an

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optimal management approach has not been established. Herein, we describe two cases of pediatric TGDC carcinoma combined with a comprehensive review of cases reported in the literature with analysis of clinical features, histopathology, and therapeutic approaches in relation to patient prognosis and outcome.

Materials and Methods

Six hundred eighty-five cases of thyroglossal duct cyst were identified from the files of the pathology departments within Southern California Permanente Medical Group hospitals between June 2005 and June 2015. The clinical data, treatment, and follow-up information were obtained from the electronic medical records augmented by the surgical pathology reports. A cohort of 22 patients had associated carcinoma, [4] with two patients <21 years at the time of presentation. Patients were retrospectively staged based on the 8th edition of the American Joint Committee on cancer staging [54], even though TGDC carcinomas are not formally included in the staging protocol. This retrospective clinical investigation was conducted in accordance and compliance with all statutes, directives, and guidelines of an Internal Review Board authorization (#5968) performed under the direction of Southern California Permanente Medical Group.

Results

Case One

A 16-year-old male presented with a chief complaint of an anterior neck mass of 3 months duration. Physical examination revealed a hard mass overlying the cricoid, which was non tender to palpation. X-ray showed an anterior superior midline neck mass at the level of the cricoid, with a computed tomography showing a cystic mass (Fig. 1). A presumptive diagnosis of a TGDC was made and a Sistrunk procedure performed. The excised cyst was 0.8 cm, with a respiratory type epithelial lining and a 0.4 cm focus of ectopic thyroid tissue within the cyst wall (Fig. 2). The cyst was associated with a 0.6 cm tumor with a papillary architecture and characteristic nuclear features of papillary thyroid carcinoma (Figs. 2, 3). The tumor extended beyond the cyst wall into the adjacent soft tissues (skeletal muscle and fat). Eight months later, a total thyroidectomy was performed without complications. Histologic examination of the thyroid gland showed no significant pathologic changes. At last follow up (111 months), the patient is alive with no evidence of recurrent or metastatic disease.

Case Two

A 12-year-old male presented with a 2-day history of a painful swelling of the anterior neck. On physical

Fig. 1 Imaging findings. Case #1: **a** A neck X-ray showing a mass at the level of the hyoid bone (marker BB). **b** A computed tomography scan shows an anterior midline cyst. Case #2: **c** Sagittal computed tomography shows an irregular soft tissue mass just below the hyoid bone. **d** A sagittal MRI T1 weighted and fat suppressed image shows the outline of the midline neck mass

Fig. 2 Histology findings. **a** A respiratory epithelial lined cyst associated with unremarkable thyroid gland tissue as part of a TGDC in case #2. **b** The upper cyst is filled with papillary carcinoma, which expands into the adjacent unremarkable thyroid gland tissue in case #1

Fig. 3 **a** The respiratory epithelial-lined cyst is immediately adjacent to a papillary carcinoma (lower field) in case #2. **b** Classical features of papillary carcinoma are present, including psammoma bodies (case #1). **c** Papillary structures and psammoma bodies in a microscopic classical papillary carcinoma (case #1)

examination, a mobile midline mass was observed in the region of the hyoid bone. Magnetic resonance imaging of the neck revealed an irregularly enhancing cystic lesion

just below the level of the hyoid (Fig. 1). The patient was prescribed a course of antibiotics with relief of his symptoms. A Sistrunk procedure was subsequently performed

without complications. The submitted specimen showed a 2.8 cm cyst, which on microscopic examination was lined by respiratory and squamous epithelium (Fig. 2). There was an associated 0.6 cm area of ectopic thyroid tissue located in the cyst wall and skeletal muscle adjacent to the cyst. Within the thyroid parenchyma, a 0.1 cm microscopic focus of papillary thyroid carcinoma was identified, characterized by follicles lined by enlarged, overlapping cells with nuclear contour irregularities, grooves, and chromatin clearing (Fig. 3). The focus of tumor involved skeletal muscle outside of the cyst wall, but did not involve the specimen margins. The patient received no further therapy and is alive without evidence of disease at last follow-up 22 months after surgery.

Discussion

Carcinomas arising from TGDCs are well described, but rare, particularly in children and adolescents. Among 685 TGDCs reviewed during a consecutive 10-year period, an associated carcinoma was identified in only two patients of pediatric age (0.3%). A review of the pertinent English literature revealed 57 cases of TGDC carcinoma occurring in patients less than 21 years of age, the vast majority of which were single case reports [4–53]. Many of these reports do not include clinical symptoms, tumor size, follow-up data, or are aggregated as part of a series report excluding individual data points. Further, Kennedy et al. [27] reported the same patient again with Branstetter et al. [55]. Joseph et al. [24] is included in many reviews as reporting pediatric patients, but the two patients are 60 and 33 years of age, respectively, and thus excluded from this review. Thus, an aggregation of the data to include all pertinent information is impossible. With these limitations in mind, the available data is summarized in Table 1.

Pediatric patients with TGDC carcinoma were slightly more common in females, with a 1.6:1 female to male ratio, different from their adult counterparts, where there is a 2.3:1 ratio [4], but also distinct from the male predominance of pediatric TGDCs in general (males:females, 1.4:1). Patients ranged in age from 6 to 20 years with an average age at presentation of 15.0 years. There was no significant difference in mean age at presentation between the sexes. All patients presented clinically with an anterior midline neck mass or swelling, which was typically mobile and painless. Two patients presented with a fixed mass [11, 42], a finding which is more suggestive of carcinoma [4] than the usual mobile mass presentation. Pain was experienced by a few patients [9, 11, 29], with an accompanying draining sinus in one patient [49]. Most patients, where the duration of symptoms was reported, had experienced symptoms for 19.0 months (average), similar to the average

Table 1 Literature summary and current cases of thyroglossal duct cyst (remnant) carcinoma in children [4–53]

Characteristics ^a	Number
Total number of patients	59
Sex	
Female	36
Male	23
Age (in years)	
Range	6–20
Mean	15.0
Female (mean)	15.1
Male (mean)	14.8
Symptom duration (in months)	
Range	0.3–96
Mean	19.0
Clinical presentation ^a	
Midline mass	59
Mobile mass	53
Fixed mass	2
Pain/tenderness	4
Fine needle aspiration	
Y:N	5:53
Papillary carcinoma	
Microscopic	11
Classical or follicular variant	37
Type/variant not stated	11
Tumor location	
Single: midline (thyroglossal duct cyst origin)	55
Multiple: midline and thyroid gland primary	4
Tumor size (cm)	
Range	8.5
Mean	2.6
Extracapsular extension (beyond thyroglossal duct cyst)	
Yes	17
No	41
Treatment	
Sistrunk only	25
Sistrunk and lymph node dissection	5
Sistrunk and thyroidectomy	14
Sistrunk, thyroidectomy and lymph node dissection	15
Radioactive iodine	20
Tumor primary group ^{a,b}	
T1	23
T1a	10
T1b	13
T2	17
T3	6
Stage I	59
Recurrence ^a	
Yes	4
No	49
Lymph node metastasis ^a	

Table 1 (continued)

Characteristics ^a	Number
Yes	11
No	47
Patient outcome (in months)	
Patients with follow up (average)	45 (55.2)
Alive with no evidence of disease (average)	40 (57.4)
Alive with evidence of disease (average)	4 (43.5)
Dead with evidence of disease	1 (13.0)

^aNot stated in all cases, ^bbased on AJCC 8th edition [54]

duration of 22 months for TGDCs in general [3]. Thus, overall, the clinical presentation is nonspecific and indistinguishable clinically from that of non-neoplastic TGDC.

Diagnostic imaging was infrequently employed in the initial evaluation of the reported patients. Ultrasound examination was most common, with lesions having a non-specific cystic appearance. Computed tomography (CT) findings reported to be of value in identifying malignancy within a TGDC, include the presence of a solid component, mural nodule, or calcifications [3, 19, 42, 43, 50, 55–72]. However, these particular imaging findings were only of assistance in a few cases [4, 38, 48, 55].

The utility of fine needle aspiration (FNA) in the preoperative evaluation of TGDC carcinoma has not been studied in the pediatric population. In adults, TGDC associated malignancies are infrequently identified by FNA, primarily due to the inherently hypocellular nature of the specimen and failure to adequately sample the malignant component, which often comprises a small area of a predominantly cystic lesion [4, 50, 73–76]. Among the reported pediatric patients with TGDC carcinoma, FNA was performed in five, and not performed in our patients [13, 37, 38, 42, 48]. One was interpreted as a thyroglossal duct cyst [13] three were atypical or suspicious for malignancy [37, 38, 42] and one was diagnosed as a papillary thyroid carcinoma [48]. While potentially specific, given the limited number of patients upon whom a FNA was performed, no specific comment about its utility can be suggested in this age group.

Macroscopically, TGDC carcinomas have a predominantly cystic appearance, although occasionally a mural nodule, solid areas, or papillary excrescences can be appreciated [8, 29, 30, 39, 46, 53]. Histologically, all reported cases of TGDC carcinomas in the pediatric population have been papillary thyroid carcinomas, a finding quite similar to adults [4]. One reported case showed concurrent transition to squamous epithelium (carcinoma) [40]. In general, the tumors are relatively large, with a mean size of 2.6 cm. The tumors showed a typical papillary and follicular architecture, with the classical nuclear

features of papillary thyroid carcinoma. The tumors were typically confined to the thyroglossal duct cyst, however extension beyond the cyst wall with involvement of surrounding pericystic soft tissue may be seen and was noted in 17 of 59 cases (29%). However, as this was not detected clinically, using the new AJCC 8th edition criteria (even though staging is not advocated), none of these tumors would be placed in a pT3 category (microscopic extra-thyroidal extension is no longer included in pT3 classification; it must be a macroscopic, clinical finding) [54].

The TGDCs are lined by respiratory and/or squamous epithelium, while ectopic thyroid gland tissue was only specifically reported in 16 cases [4, 11, 15, 19, 25, 28–30, 33, 37, 40, 41, 49, 52]. By extension, however, the presence of a thyroid follicular based neoplasm, suggests thyroid gland tissue origin. Further, as carcinoma was only identified in four patients who also had a thyroidectomy [10, 37, 39, 43] it seems to confirm a primary origin from ectopic thyroid gland tissue rather than representing metastatic disease. Other considerations would include a papillary thyroid carcinoma arising in the pyramidal lobe of the thyroid gland or metastatic carcinoma to the Delphian lymph node, but these considerations can be easily excluded by clinical and pathology findings. In the present cases, no association with the thyroid gland was identified (pyramidal lobe), and no lymph node tissue was present.

All patients were managed surgically, with a Sistrunk procedure performed in all cases. This approach adequately manages both TGDC as well as cases that may harbor a clinically silent or recognized carcinoma. This procedure results in an excellent long term outcome without any additional complications [3, 4, 77, 78]. It would seem that with such a low chance of a concurrent primary thyroid gland tumor (all primary thyroid gland tumors were microscopic tumors and thus would not have been clinically detected [10, 37, 39, 43]), the 6.6% chance of detecting an indolent or microscopic thyroid gland primary would make this procedure unnecessary. Conservative excision with clinical follow-up would be prudent, in an effort at de-escalation therapies for tumors which are quite indolent. Only a single patient died with disease, a patient reported from 1959, [26] where she was followed for 7 years before surgery and never had a thyroidectomy, thus precluding the identification of a thyroid gland primary, which may have resulted in the disseminated disease to lung and liver (identified at autopsy).

If follow-up or concurrent thyroidectomy is not performed, then radioablative iodine therapy is not employed. In the reported cases, radioactive iodine was used for patients with lymph node metastases (n=6) [5, 10, 16, 35, 42], but also used as ablation of any potential thyroid remnant.

In pediatric patients, all tumors are considered Group I by definition, unless there is distant disease at presentation [54]. However, staging for TGDC carcinomas is presently not advocated nor included in the 8th edition of the AJCC staging manual [54]. Two points for consideration: soft tissue extension of TGDC carcinomas could be construed as “extra thyroidal” extension, but since this criteria has been removed as a microscopic criteria for pT-group determination, this would not be a consideration in this anatomic site either. The second point is that even with lymph node metastases, all cases, if staged, would be Group I tumors, a group that generally enjoys an excellent long term prognosis. Further, lymph node metastases were identified in only four patients. To wit, in this review of the literature, only a single patient died of disease (1.6%), a finding that would match current differentiated thyroid carcinoma outcome when identified as a thyroid gland primary [63]. Disease persistence/recurrence was observed in only four patients [10, 11, 35], but follow-up was on-going at the time of report, so they may have been adequately managed. While the number of patients is small, outcome did not appear to be influenced by tumor invasion of soft tissues, presence of a synchronous thyroid gland carcinoma, or use of adjunctive therapies.

Conclusion

Carcinomas associated with TGDCs are exceedingly rare in the pediatric population. Patients are more commonly female, typically exhibiting a midline cervical mass indistinguishable clinically from a benign TGDC. Histologically, all TGDC associated malignancies in this age group have been papillary thyroid carcinomas. All patients presented as stage I tumors. Persistent/recurrent disease is seen in a minority of patients, with a very low risk of distant metastasis. The Sistrunk procedure is considered to be the treatment of choice. Given the excellent clinical outcome observed in most pediatric cases of TGDC carcinoma, additional surgery and/or radioablative iodine seem to yield no outcome advantage. Additional evaluation of larger cohorts of patients should aid in confirming this recommendation.

Compliance with Ethical Standards

Conflict of interest All authors declare there are no conflicts of interest. The opinions or assertions contained herein are the private views of the author and are not to be construed as official or as reflecting the views of Southern California Permanente Medical Group.

Ethical Approval All research was retrospective involving human participants in accordance with the ethical standards of the institution with IRB approval #5968.

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