Head and Neck Pathol
DOI 10.1007/s12105-016-0724-7

ORIGINAL PAPER

A Clinicopathologic Series of 685 Thyroglossal Duct Remnant Cysts

Lester D. R. Thompson[1] · **Hannah B. Herrera**[1] · **Sean K. Lau**[2]

Received: 29 March 2016 / Accepted: 27 April 2016

Abstract The clinical features of thyroglossal duct remnant cysts (TGDC) have been well described, however the histopathologic aspects of these lesions have not been addressed in a detailed manner. In particular, there has been no large community practice based series evaluating TGDC histologically compared with management outcomes. A retrospective review of all TGDC diagnosed between 2005 and 2015 was performed. Six hundred eighty-five patients were identified (344 males; 341 females). Age at presentation was bimodal (first and fifth decades) and ranged from 0.8 to 87 years (mean 31.3 years). Males predominate in children (150:111); females in adults (230:194). Patients presented most frequently with a mobile midline neck mass in an infrahyoid location. An associated skin fistula (n = 67) was twice as common in pediatric as adult patients. The average cyst size was 2.4 cm (range 0.4–9.9 cm) by imaging studies and 2.6 cm (range 0.2–8.5 cm) by pathologic examination; pediatric patients had smaller cysts (mean 2.1 cm) than adults (mean 2.8 cm). Histologically, 257 (38 %) TGDC were lined by respiratory epithelium alone, 68 (10 %) squamous epithelium alone, 347 (51 %) exhibited both respiratory and squamous epithelium, and 13 (1 %) had no identifiable epithelial lining. Four hundred eighty-four (71 %) TGDC had associated thyroid gland tissue present within the cyst wall (n = 282), skeletal muscle (n = 71), adipose tissue (n = 34), or a combination of these sites (n = 97). The hyoid bone was identified in 647 (grossly and/or histologically), and absent in 38. Surgical management consisted of Sistrunk procedure (n = 647), cystectomy (n = 31), or thyroidectomy/thyroid lobectomy (n = 7). Treatment related complications were observed in 6 patients, which included vocal cord damage, seroma, and hematoma. Recurrences developed in 20 (3 %) patients, 14 of whom were managed initially by cystectomy. Papillary thyroid carcinoma was identified in 22 (3.2 %) TGDC. In summary, TGDC show a bimodal peak in the 1st and 5th decades, commonly presenting as a midline cervical lesion below the hyoid bone, associated with a skin fistula in 10 %. Histologically TGDC are most commonly lined by a combination of respiratory and squamous epithelium. Thyroid gland tissue is identified in 71 % of cases (0.45 cm mean size), although not limited to the cyst wall, but present in the surrounding soft tissues. Rare TGDC may harbor malignancy (3.2 %). TGDC are most effectively managed by Sistrunk procedure rather than excision, which carries low rates of complications (1 %) and recurrence (3 %).

Keywords Thyroglossal cyst · Respiratory mucosa · Retrospective studies · Recurrence · Humans · Adult · Child · Thyroid neoplasms · Cutaneous fistula · Cystectomy

The opinions or assertions contained herein are the private views of the authors and are not to be construed as official or as reflecting the views of Southern California Permanente Medical Group.

✉ Sean K. Lau
Sean.K.Lau@kp.org

[1] Department of Pathology, Woodland Hills Medical Center, Southern California Permanente Medical Group, 5601 De Soto Avenue, Woodland Hills, CA 91364, USA

[2] Department of Pathology, Orange County-Anaheim Medical Center, Southern California Permanente Medical Group, 3440 East La Palma Avenue, Anaheim, CA 92806, USA

Introduction

The thyroglossal duct originates during embryonic development of the thyroid gland. As the thyroid anlage descends caudally in the neck, it is thought to form a duct that

Published online: 09 May 2016

remains connected to its point of origin at the level of the foramen cecum of the tongue [1–5]. The thyroglossal duct typically involutes and atrophies between 7 and 10 weeks gestation following migration of the primitive thyroid to its final pretracheal position in the inferior neck. Vestiges of the inferior thyroglossal duct are not uncommon, forming the pyramidal lobe of the thyroid gland. Persistence of other portions of the thyroglossal duct may give rise to cysts.

Thyroglossal duct remnant cysts (TGDC) are among the most common cervical lesions encountered in infancy and childhood [6, 7], although they not infrequently present in adults [8–13]. The cysts may arise anywhere along the migratory pathway of the thyroglossal duct of the developing thyroid gland. Histologically, TGDC are lined by respiratory epithelium, squamous epithelium, or a combination of both. Microscopic foci of ectopic thyroid tissue are variably present [2–4].

Although multiple studies have addressed the clinical features of TGDC, few have included a detailed analysis of the associated pathologic findings [8, 13–20]. To date, there has been no large community practice based series evaluating TGDC histologically compared with management outcomes.

Materials and Methods

Six hundred eighty-five cases of TGDC were identified from the files of the Departments of Pathology within Southern California Permanente Medical Group between June 2005 and June 2015. Hematoxylin and eosin stained slides from all cases were reviewed, with a range of 1–34 slides (mean 3 slides) per case available for analysis. Clinical data, treatment, and follow-up information were obtained from electronic medical records. Seven hundred eleven cases were initially reviewed, with 26 cases reclassified and excluded from the study: 10 dermoid cysts, 9 foregut duplication cysts, and 7 cutaneous epidermoid cysts. This 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

Clinical Features

The study population included 344 males and 341 females, with an overall mean age at presentation of 31.3 years (range 0.8–87 years) (Table 1). A bimodal age peak was

Table 1 Clinical characteristics of study population

Characteristics[a]	Number
Total number of patients	685
Gender	
Male	344
Female	341
Age (in years)	
Range	0.8–87
Mean	31.3
Age deciles	
0–9	179
10–19	82
20–29	63
30–39	92
40–49	103
50–59	83
60–69	56
70–79	24
80–89	3
Age 0–19: Males: Females	150:111
Age 20–87: Males: Females	194:230
Symptom duration (in months)	
Range	0.2–456
Mean	22
Symptom duration for carcinoma cases	14.5
Clinical presentation	
Mobile mass	680
Fixed mass	5
Pain/tenderness	164
Infection	123
Drainage or fistula	67
Imaging size (cm)	
Range	0.4–9.9
Mean	2.4
Pathologic size (cm)	
Range	0.2–8.5
Mean	2.6
Laterality	
Midline	668
Left	9
Right	8
Location	
Suprahyoid	165
Infrahyoid	520
Treatment	
Sistrunk	624
Sistrunk and thyroidectomy	22
Sistrunk and thyroid lobectomy	1
Cystectomy	31
Thyroidectomy	3

Table 1 continued

Characteristics[a]	Number
Thyroid lobectomy	4
Complications	6
Recurrence	20

[a] Data parameters listed were not stated in all cases

identified with 179 (26 %) patients in the first decade, while 103 (15 %) patients were in the 5th decade. However, in adults, patients were most frequently affected between the fourth and sixth decades. Among children there was a male preponderance (150 males: 111 females; 1.4:1), with a female preponderance in adults (194 males: 230 females; 1:1.2). Of the 685 patients, 519 were White, 87 were Black, and 79 were Asian.

In relation to the hyoid bone, 520 (76 %) TGDC were infrahyoid and 165 (24 %) were suprahyoid. The most common clinical presentation was a midline neck mass (Fig. 1a), observed in 668 (98 %) patients. All of the submitted cases, including the excluded cases, were submitted as TGDC clinically. A minority presented with a lateral neck mass either to the right (n = 8) or left (n = 9) of midline. The mass was typically mobile (n = 683) and nontender (n = 522). Five patients exhibited a fixed mass; of these three had thyroid carcinomas associated with the TGDC. Secondary clinical infection of the cyst was reported in 123 (18 %) patients. Sixty-seven (9.8 %) patients presented with a discharging cutaneous fistula. Difficulty swallowing was reported in 33 (4.8 %) patients. The duration of symptoms ranged from 0.2 to 456 months with a mean of 22 months. However, the symptom duration was much shorter in patients who had carcinoma (mean 14.5 months).

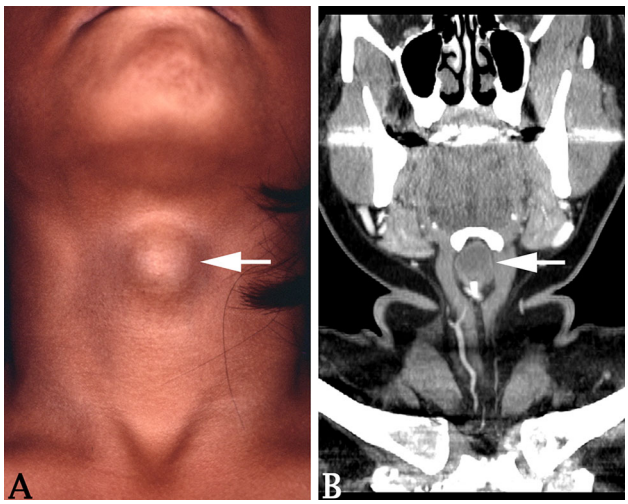

Fig. 1 Clinical manifestations of thyroglossal duct remnant cysts. **a** An infrahyoid midline neck mass (*arrow*), which was soft and mobile with tongue protrusion. **b** A computed tomography scan showing a midline cystic mass (*arrow*) immediately below the hyoid bone

Radiographic assessment was performed preoperatively in 703 patients. Imaging studies included computed tomography (CT) in 383 patients (Fig. 1b), ultrasound (US) in 275 patients, magnetic resonance imaging (MRI) in 39 patients, and radioisotope thyroid scanning in 6 patients. Two or more studies were performed in 92 patients, the majority of whom underwent US and CT. By imaging studies, TGDC ranged from 0.4 to 9.9 cm in greatest dimension, with an average size of 2.4 cm.

Preoperative fine needle aspiration (FNA) was performed in 144 patients. Utilizing the Bethesda System for reporting thyroid cytopathology, the aspirates were classified as follows: nondiagnostic or unsatisfactory (n = 123), benign (n = 13), atypia of undetermined significance or follicular lesion of undetermined significance (n = 2), follicular neoplasm or suspicious for follicular neoplasm (n = 1), suspicious for malignancy (n = 2), malignant (n = 3). All of the Category IV (follicular neoplasm), Category V (suspicious for malignancy), and Category VI (malignant) cases (n = 6), correlated correctly with TGDC associated papillary carcinoma (excellent predictive values). Twelve of the 22 carcinoma cases had preoperative FNAs; the remaining cases that had FNAs were called: Category II (n = 4) and Category I (n = 2). Thus, it seems that FNA performed on firm, fixed lesions may yield a meaningful preoperative finding of carcinoma, while the vast majority are non-diagnostic, and thus, perhaps unnecessary.

Pathologic Features

On gross examination TGDC ranged from 0.2 to 8.5 cm in greatest dimension (mean 2.6 cm). The cysts in pediatric patients were smaller (mean 2.1 cm) than those of adult patients (mean 2.8 cm). Two hundred fifty-seven (38 %) TGDC were lined by respiratory epithelium only, 68 (10 %) were lined by squamous epithelium only, and 347 (51 %) were lined by a combination of respiratory and squamous epithelium (Table 2; Fig. 2). The respiratory epithelium had a variable appearance, comprised of pseudostratified ciliated columnar or low cuboidal cells. The squamous epithelium was stratified and predominantly nonkeratinizing. Keratinization was observed in four cases. Subepithelial inflammation was present in 596 (87 %) TGDC (Fig. 3). In five cases, there was prominent lymphoid hyperplasia characterized by numerous lymphoid follicles containing germinal centers. In 443 (65 %) cases, there was microscopic evidence of cyst rupture associated with foamy histiocytes (Fig. 3b), cholesterol clefts, and a foreign body giant cell reaction. Thirteen (1 %) TGDC

Table 2 Pathologic features of thyroglossal duct remnant cysts

Characteristics	Number
Respiratory epithelium only	257
Squamous epithelium only	68
Respiratory and squamous epithelium	347
No epithelial lining	13
Inflammation	596
Mucoserous glands	104
Cartilage within cyst wall	5
Hyoid bone	647
Thyroid gland tissue identified	484
Cyst wall only	282
Cyst wall and skeletal muscle	44
Cyst wall and adipose tissue	49
Skeletal muscle only	71
Adipose tissue only	34
Skeletal muscle and adipose tissue	4
Papillary thyroid carcinoma	22

lacked any identifiable epithelial lining, which was replaced by fibroinflammatory tissue (Fig. 4).

Ectopic thyroid gland tissue was characterized by groups of colloid filled follicles identified in 484 (71 %) specimens. The thyroid gland tissue ranged from 0.01 to 5.0 cm (mean 0.45 cm) and was localized to the cyst wall (n = 282), skeletal muscle (n = 71), adipose tissue (n = 34), or a combination of these sites (Figs. 2, 3, 4, 5). Curiously, eleven (1.6 %) TGDC were identified within the thyroid gland, involving the left lobe (n = 4), right lobe (n = 1), isthmus (n = 1), and pyramidal lobe (n = 5). However, the cyst was the dominant finding by gross measurement. A thyroid gland carcinoma was identified in 22 (3.2 %) TGDC. All were classic papillary thyroid carcinomas, ranging in size from 0.1 to 3.8 cm (mean 1.4 cm), exhibiting characteristic nuclear features including enlargement, overlapping, irregular contours, clear chromatin, grooves, and pseudoinclusions. Other histologic findings associated with TGDC included the presence of mucoserous salivary gland tissue (n = 104; Fig. 4d) and foci of cartilage within the cyst wall (n = 5). Hyoid bone was present in 647 specimens (Fig. 5), with microscopic examination of the osseous tissue revealing no pathologic abnormalities.

Treatment and Follow-Up

Six hundred twenty-four patients were treated with a Sistrunk procedure. Named after Dr. Walter E. Sistrunk (1880–1933) who developed the technique in the 1920's, the surgery

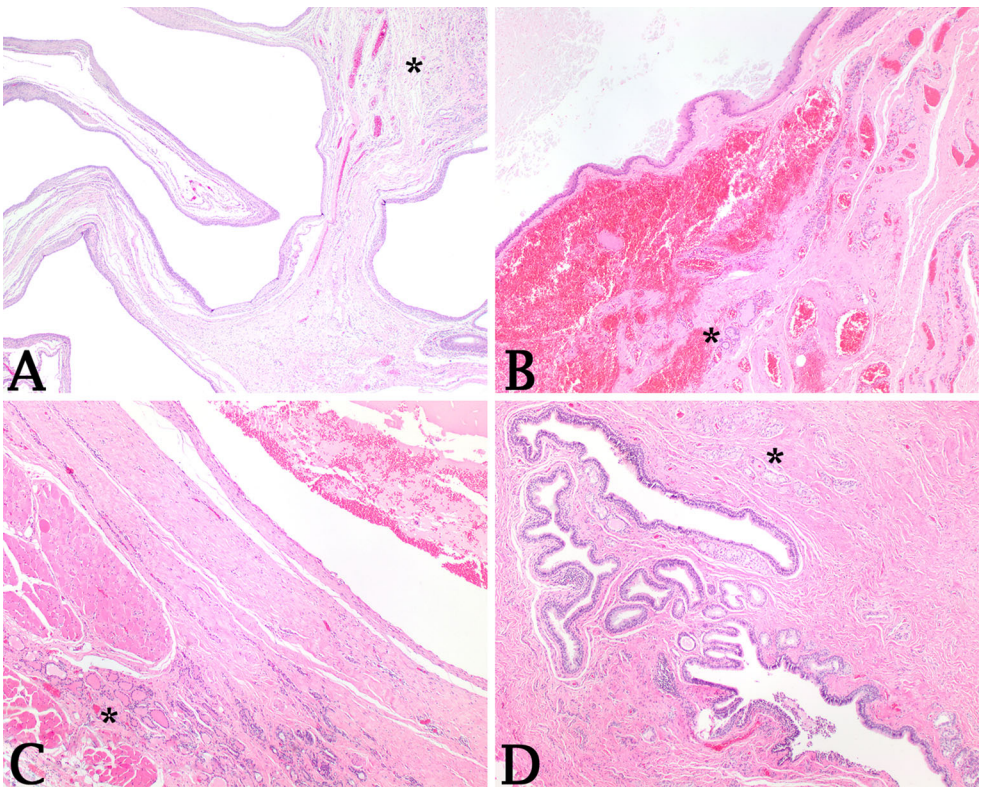

Fig. 2 Thyroglossal duct cyst lining. **a** Both respiratory and squamous epithelium are noted lining the multiple cystic spaces. **b** A squamous epithelium lines the cyst. **c** The epithelium is attenuated and absent. **d** Ciliated respiratory type epithelium is seen within the cystic spaces. Thyroid gland tissue (*asterisk in each image*) is noted within the cyst wall and adjacent skeletal muscle

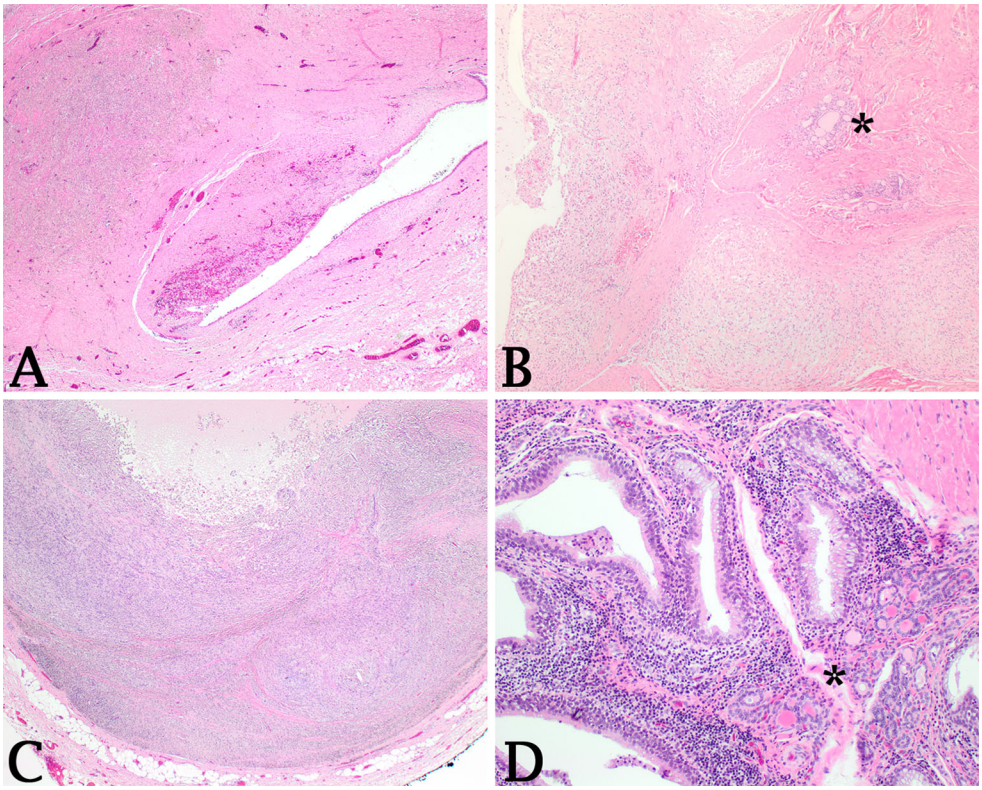

Fig. 3 Inflammation is a common finding in thyroglossal duct cysts. **a** Hemosiderin laden macrophages are noted in the cyst wall. **b** Collections of histiocytes are noted within the cyst and in the stroma. **c** The cyst has been nearly completely replaced by reactive histiocytes and fibroblasts. **d** A prominent lymphoid infiltrate is noted around the cystic spaces, intermingled with the thyroid gland tissue (*asterisk in each image*)

involves the removal en bloc of the cyst, fistula, middle third of the hyoid bone, and a core of tissue to the base of the tongue. A Sistrunk procedure with concurrent thyroidectomy or thyroid lobectomy was performed in 23 patients. Thirty-one patients underwent cystectomy without excision of the hyoid bone. A subset of TGDC localized to the thyroid gland were managed by thyroidectomy (n = 3) or thyroid lobectomy (n = 4) alone. Treatment associated complications included vocal cord damage (n = 1), seroma (n = 3) and postoperative bleeding (n = 2). Clinical follow up information was available for 678 patients with a mean duration of 46 months (range 0.1–136 months). There were 20 recurrences among the 685 patients for an overall recurrence rate of 3 %. The group of patients with recurrences had a mean age of 19 years (range 1–45 years). The average time interval to recurrence was 19.5 months (range 1–72 months). Of the 20 recurrences, 14 occurred following cystectomy, with the remaining 6 following Sistrunk procedure.

Discussion

The thyroid gland originates as an invagination of proliferating endodermal cells in the floor of the pharynx during the fourth week of embryonic development [1–5]. As the thyroid anlage descends, it maintains an attachment to the site of what will become the foramen cecum known as the thyroglossal duct. The thyroglossal duct atrophies and disappears between the 7th and 10th weeks of gestation. Persistent remnants of the thyroglossal duct are thought to be the source of TGDC. The exact incidence of thyroglossal duct remnants is unknown. Microscopic examination of 200 adult larynges reported by Ellis and van Nostrand showed thyroglossal duct remnants in 7 % of specimens [21], while an autopsy study involving 58 pediatric patients identified remnants of the thyroglossal duct or ectopic thyroid tissue in 41 % of cases [17]. Clinical manifestation of TGDC based on a mid-year mean patient population of 3,064,661 patients for Southern California Permanente Medical Group would correspond to 2.23/100,000 population at risk each year.

In our series, TGDC showed no gender predilection, a finding similar to the literature [3, 8–13]. However, males tended to predominate in pediatric patients while females predominated in adults. There is a wide age range at presentation. While TGDC have been reported predominantly in the pediatric population, there appears to be a bimodal distribution of disease as evidenced in our patient cohort (1st and 5th decades) and other studies which have included both children and adults [3, 10–12].

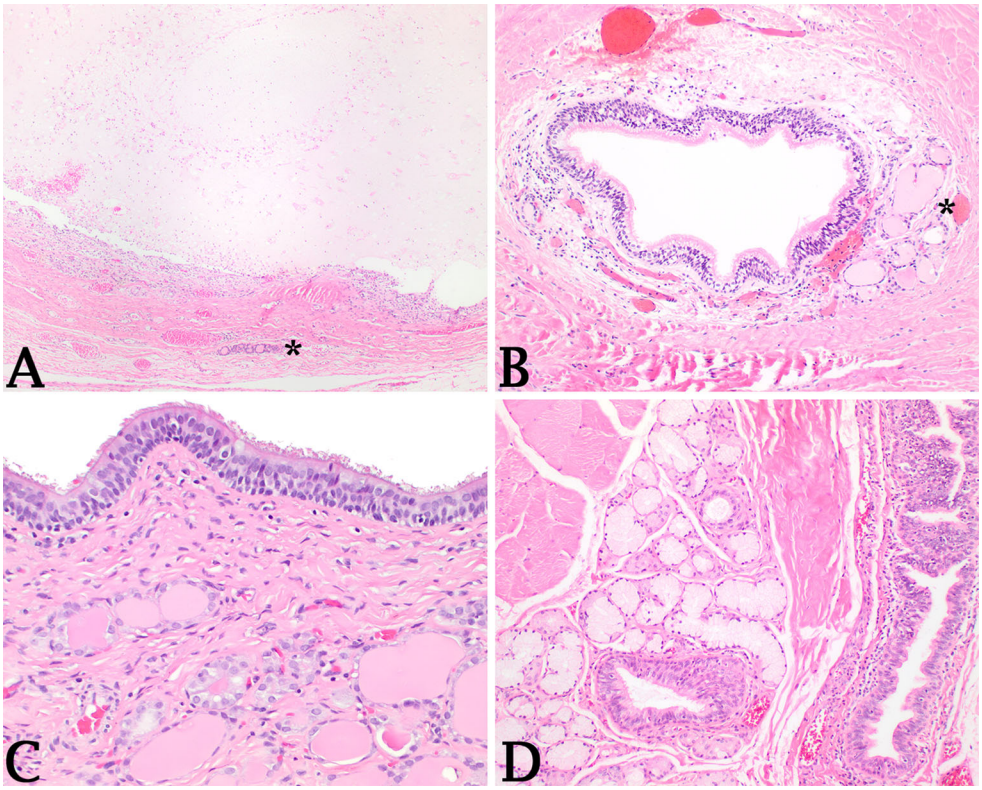

Fig. 4 The amount of thyroid gland tissue (*asterisk in each image*) present as well as the location of the tissue was quite variable. **a** A collection of a few thyroid follicles in the cyst wall. **b** A few thyroid follicles adjacent to a respiratory epithelial lined cyst. **c** Easily identified thyroid tissue with colloid in the cyst wall. **d** Minor mucoserous glands noted in association with the cyst and with skeletal muscle

The classic clinical presentation of TGDC is a mobile painless mass in the midline of the neck. Rarely, the cysts may be located laterally [8, 10–13, 15, 22], as observed in 2 % of the patients in the present series. Infection of the cyst is also a common presentation, frequently accompanied by a discharging fistula. A cutaneous fistula was observed in 10 % of the patients in the current study, and was twice as common in pediatric as adult patients. This differs from data reported by others indicating this particular clinical presentation to be significantly more frequent in adults than in children [12, 23]. TGDC are uncommonly associated with dysphagia or airway obstruction [10, 12, 13, 24]. A recent meta analysis of 1015 patients with cystic cervical masses, highlighted infection/abscess, fistula/draining sinus, dysphagia, and airway obstruction as the main clinical presentation and symptoms of TGDC in decreasing order of frequency [25].

TGDC can arise anywhere along the migratory path of the developing thyroid gland. The vertical location is conventionally described in relation to the hyoid bone, with the vast majority occurring at the level of or inferior to the hyoid bone. The overall distribution of TGDC in the present study was similar to that documented by others, with approximately three quarters of cysts positioned in an infrahyoid location [3, 8–11, 13, 15, 22, 24]. TGDC can rarely occur in the tongue, mediastinum, or larynx [3, 4, 26]. Although not emphasized in prior large series, TGDC may also arise within the substance of the thyroid gland, as evidenced by the eleven cases of intrathyroidal TGDC observed in the current study. FNA performed on firm, fixed lesions may yield a meaningful preoperative finding of carcinoma, while the vast majority are non-diagnostic, and thus, perhaps unnecessary.

Histologically, TGDC are characteristically lined by respiratory (columnar to stratified cuboidal) and/or squamous epithelium, though the relative frequency of these types of epithelia have only rarely been documented [8, 13–16, 19, 27]. We found a lining comprised of both respiratory and squamous epithelium to be the most common (51 %), followed by respiratory epithelium (38 %) and squamous epithelium (10 %) alone. TGDC frequently show evidence of an inflammatory infiltrate, but this fails to correlate with clinical evidence of infection. Marked inflammation of the cyst can lead to destruction of the epithelial lining and replacement by fibroinflammatory or granulation tissue. Inflammation may also result in disruption of the cyst wall with the development of a foreign body giant cell reaction. In many cases, the inflammation

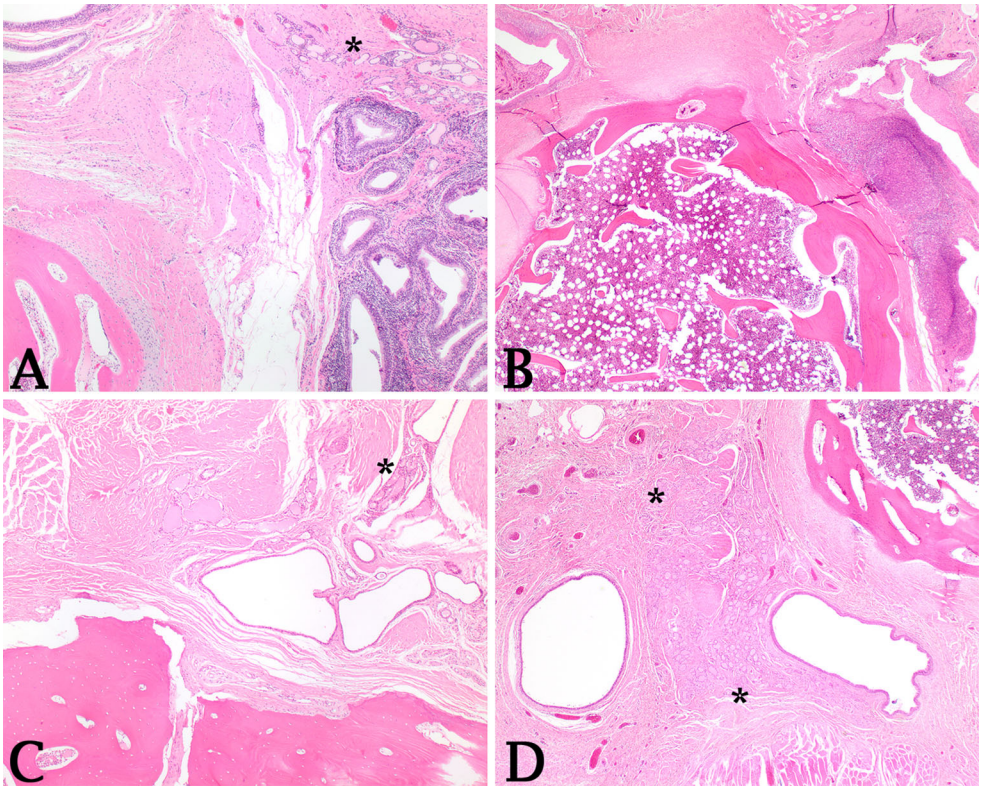

Fig. 5 While the presence of the hyoid bone did not disclose any microscopic pathology, it did help to orient the specimen and suggest the location of the cyst. **a** Multilocular cyst with inflammation and thyroid (*asterisk in each image*) tissue and bone. **b** Cellular marrow within the hyoid bone, adjacent to a squamous lined cyst with germinal center formation. **c** Bone adjacent to thyroid gland tissue and cysts. **d** Ample thyroid tissue next to epithelial lined cyst and bony tissue

may be the dominant finding, requiring careful high power examination to document the cyst lining or the ectopic thyroid gland tissue. Rarely reported features include the presence of mucoserous salivary glands and foci of cartilage within the cyst wall [2, 14]. The separation of a bronchogenic cyst from a TGDC may be difficult in these circumstances, unless the thyroid gland tissue can be identified. Further, the presence of smooth muscle is usually only seen in bronchogenic cysts and not in TGDC. The reported incidence of ectopic thyroid tissue associated with TGDC varies among different studies [13–17, 20, 27] but has been reported from 31 up to 62 % of cases [28]. In the current study, thyroid gland tissue was identified in 71 % of the specimens based on routine examination and tissue sampling (mean 3 slides per case). The high frequency of thyroid tissue identification in this study may be as a result of primary care review, rather than the referral bias seen in academic institutions, particularly towards malignancy [20]. While the thyroid gland tissue was present most frequently in the cyst wall, it was also noted within adipose tissue and skeletal muscle adjacent to the cyst. Thus, examination of the pericystic tissues must be done to document the thyroid gland tissue. Further, with thyroid gland tissue noted in the skeletal muscle and soft tissues, if a carcinoma arises from these ectopic tissues, the carcinoma will show soft tissue involvement by default, rather than representing soft tissue extension.

In rare instances, TGDC may harbor malignancy. The reported incidence of carcinoma occurring in TGDC is around 1 % [13, 28–30], though higher rates have been reported by tertiary or academic referral centers, ranging from 4.9 to 7.4 % [20, 31, 32]. In the present community practice based series, 3.2 % of TGDC contained an associated carcinoma. Most thyroglossal duct cyst carcinomas are clinically indistinguishable from TGDC, with the diagnosis established only after pathologic examination of the excised specimen [3]. However, a sudden increase in size or clinical presentation of a fixed, hard mass may lead one to suspect malignancy [4, 28]. Histologically the vast majority (>99 %) of thyroglossal duct carcinomas are papillary thyroid carcinomas, with mixed papillary/follicular carcinoma, squamous cell carcinoma, and other histologic types of thyroid epithelial malignancies considered extraordinary [3, 4]. All TGDC associated malignancies in the present series were papillary thyroid carcinomas, with no other carcinoma types encountered. The cytomorphonuclear features are

classical for papillary thyroid carcinoma. Involvement of skeletal muscle may be seen, and should perhaps suggest upstaging to a pT3 tumor. Histologic examination of the thyroid in patients with thyroglossal duct cyst carcinoma may show a microscopic papillary carcinoma in approximately one-third of cases [29–33]. Among patients with thyroglossal duct cyst carcinoma who had a thyroidectomy in the present study, 32 % showed a synchronous papillary thyroid carcinoma. There is a theoretic consideration of an intrathyroidal papillary thyroid carcinoma metastasizing to a thyroglossal duct cyst. This distinction requires careful clinical and imaging correlation. The low incidence of thyroid gland involvement suggests total thyroidectomy may not be necessary for management of patients with TGDC malignancies.

In the absence of thyroid gland tissue, the histologic appearance of TGDC can be confused with other cystic lesions which may be encountered in the neck. However, it must first be stated that very careful review of all of the tissue removed may be necessary to identify the very small (mean 0.45 cm) amount of thyroid gland tissue. Branchial cleft cysts, similar to TGDC are lined by nonkeratinizing stratified squamous or columnar respiratory type epithelium. The lining epithelium is characteristically accompanied by a dense, nodular lymphoid infiltrate in the majority of cases. In contrast, lymphoid aggregates are a rare finding in TGDC, as shown in the present series. Branchial cleft cysts are encountered in the lateral neck along the anterior border of the sternocleidomastoid muscle in contrast to the typical midline clinical presentation of TGDC [6, 7].

Similar to TGDC, dermoid cysts tend to occur along the midline neck [6]. Microscopically, dermoid cysts show a lining of keratinizing squamous epithelium with hair follicles and supporting adnexa. The presence of pilosebaceous structures serves to differentiate dermoid cysts from TGDC. In addition, as shown in the present study, the lining squamous epithelium of TGDC is nonkeratinizing in all but rare cases. Sometimes the separation of a pilosebaceous unit from minor mucoserous glands may be difficult in limited material. Additional tissue or serial sections usually helps to resolve this issue.

Epidermal inclusion cysts can present as an anterior neck mass, which may be mistaken for TGDC [34]. Epidermoid cysts have a wall lined by keratin producing squamous epithelium, which differs from the squamous lining of TGDC that in most instances is nonkeratinizing. Clinically, epidermoid cysts are situated within the dermis or subcutaneous tissues (superficial) while TGDC tend to be localized in the deeper soft tissues with extension in and around the hyoid bone.

Due to the presence of a respiratory type epithelial lining, TGDC bear some resemblance to a bronchogenic cyst. Although typically localized to the anterior mediastinum, cervical examples of bronchogenic cysts have been described [35, 36]. The cyst wall of bronchogenic cysts typically contains smooth muscle, mucoserous glands, and often cartilaginous tissue. TGDC may infrequently contain submucosal glands or ectopic cartilage, but as a rule do not exhibit a muscular wall.

TGDC may rarely occur within the substance of the thyroid gland [37, 38]. An intrathyroidal presentation was observed in eleven patients in the present study. The histologic appearance of intrathyroidal TGDC overlaps with that of lesions previously described as lymphoepithelial cysts or branchial cleft like cysts of the thyroid gland [39–41]. Unlike TGDC, intrathyroidal lymphoepithelial cysts are lined predominantly by squamous epithelium. Respiratory type epithelium may be intermixed, but is typically absent or only a focal finding [38]. The presence of a dense nodular or diffuse lymphoid infiltrate within the cyst wall is a constant feature. In contrast, abundant lymphoid tissue is only rarely seen in TGDC.

Complications associated with TGDC surgery are rare. A recent review reported a complication rate of 8 % [25]. Most complications are minor, and carry minimal morbidity including local infection, seroma, hematoma, and wound dehiscence [4, 10, 16, 24, 25]. In reviews of the published literature, the overall reported risk of recurrence following surgical treatment of TGDC is approximately 7.3–11 % [24, 25, 42]. The standard treatment for TGDC is the Sistrunk procedure. This ensures removal of the entire thyroglossal duct remnants as the procedure includes removal of the mid portion of the hyoid bone along with a cylinder of tissue to include the base of tongue. Incomplete surgical removal has the greatest impact on recurrence as recurrence rates have been shown to be significantly higher in patients managed by cystectomy rather than Sistrunk procedure [24, 25, 42]. In this clinical series, 70 % of recurrences occurred in patients treated by cystectomy, further emphasizing the importance of the Sistrunk operation in the surgical management of TGDC.

To our knowledge, this study represents the largest series of TGDC described in the literature to date. TGDC have a bimodal age distribution and affect the genders equally. The lesion manifests principally as a midline cervical neck mass and is most commonly localized inferior to the hyoid bone. Histologically, TGDC are characterized most frequently by a lining composed of both respiratory and squamous epithelium. Associated ectopic thyroid gland parenchyma is present in most cases, and occurs predominantly within the wall of the cyst, but may also be present in the surrounding soft tissues. TGDC carcinomas are uncommon, with all accounted for by papillary thyroid carcinoma. TGDC are best managed

surgically by the Sistrunk procedure, which carries low rates of complications (1 %) and recurrence (3 %).

References

1. Brintnall ES, Davies J, Huffman WC, Lierle DM. Thyroglossal ducts and cysts. AMA Arch Otolaryngol. 1954;59(3):282–9.
2. Mandell L, Baurmash H. Thyroglossal tract abnormalities. Oral Surg Oral Med Oral Pathol. 1957;10(11):1131–44.
3. Allard RH. The thyroglossal cyst. Head Neck Surg. 1982;5(2):134–46.
4. Mondin V, Ferlito A, Muzzi E, Silver CE, Fagan JJ, Devaney KO, Rinaldo A. Thyroglossal duct cyst: personal experience and literature review. Auris Nasus Larynx. 2008;35(1):11–25.
5. Chou J, Walters A, Hage R, Zurada A, Michalak M, Tubbs RS, Loukas M. Thyroglossal duct cysts: anatomy, embryology and treatment. Surg Radiol Anat. 2013;35(10):875–81.
6. Koeller KK, Alamo L, Adair CF, Smirniotopoulos JG. Congenital cystic masses of the neck: radiologic-pathologic correlation. Radiographics. 1999;19(1):121–46 **quiz 152-3**.
7. Nicollas R, Guelfucci B, Roman S, Triglia JM. Congenital cysts and fistulas of the neck. Int J Pediatr Otorhinolaryngol. 2000;55(2):117–24.
8. Kuroda T, Iwasa T, Miyakawa M, Makiuchi M, Furihata R. Clinicopathological studies on thyroglossal duct remnant. Jpn J Surg. 1979;9(1):32–6.
9. Katz AD, Hachigian M. Thyroglossal duct cysts. A thirty year experience with emphasis on occurrence in older patients. Am J Surg. 1988;155(6):741–4.
10. Brousseau VJ, Solares CA, Xu M, Krakovitz P, Koltai PJ. Thyroglossal duct cysts: presentation and management in children versus adults. Int J Pediatr Otorhinolaryngol. 2003;67(12):1285–90.
11. Lin ST, Tseng FY, Hsu CJ, Yeh TH, Chen YS. Thyroglossal duct cyst: a comparison between children and adults. Am J Otolaryngol. 2008;29(2):83–7.
12. Ren W, Zhi K, Zhao L, Gao L. Presentations and management of thyroglossal duct cyst in children versus adults: a review of 106 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2011;111(2):e1–6.
13. de Tristan J, Zenk J, Künzel J, Psychogios G, Iro H. Thyroglossal duct cysts: 20 years' experience (1992–2011). Eur Arch Otorhinolaryngol. 2015;272(9):2513–9.
14. Sade J, Rosen G. Thyroglossal cysts and tracts. A histological and histochemical study. Ann Otol Rhinol Laryngol. 1968;77(1):139–45.
15. Hawkins DB, Jacobsen BE, Klatt EC. Cysts of the thyroglossal duct. Laryngoscope. 1982;92(11):1254–8.
16. Hoffman MA, Schuster SR. Thyroglossal duct remnants in infants and children: reevaluation of histopathology and methods for resection. Ann Otol Rhinol Laryngol. 1988;97(5 Pt 1):483–6.
17. Sprinzl GM, Koebke J, Wimmers-Klick J, Eckel HE, Thumfart WF. Morphology of the human thyroglossal tract: a histologic and macroscopic study in infants and children. Ann Otol Rhinol Laryngol. 2000;109(12 Pt 1):1135–9.
18. Chandra RK, Maddalozzo J, Kovarik P. Histological characterization of the thyroglossal tract: implications for surgical management. Laryngoscope. 2001;111(6):1002–5.
19. Ali AA, Al-Jandan B, Suresh CS, Subaei A. The relationship between the location of thyroglossal duct cysts and the epithelial lining. Head Neck Pathol. 2013;7(1):50–3.
20. Wei S, LiVolsi VA, Baloch ZW. Pathology of thyroglossal duct: an institutional experience. Endocr Pathol. 2015;26(1):75–9.
21. Ellis PD, van Nostrand AW. The applied anatomy of thyroglossal tract remnants. Laryngoscope. 1977;87(5 Pt 1):765–70.
22. Yaman H, Durmaz A, Arslan HH, Ozcan A, Karahatay S, Gerek M. Thyroglossal duct cysts: evaluation and treatment of 49 cases. B-ENT. 2011;7(4):267–71.
23. Pradeep PV, Jayashree B. Thyroglossal cysts in a pediatric population: apparent differences from adult thyroglossal cysts. Ann Saudi Med. 2013;33(1):45–8.
24. Rohof D, Honings J, Theunisse HJ, Schutte HW, van den Hoogen FJ, van den Broek GB, Takes RP, Wijnen MH, Marres HA. Recurrences after thyroglossal duct cyst surgery: results in 207 consecutive cases and review of the literature. Head Neck. 2015;37(12):1699–704.
25. Gioacchini FM, Alicandri-Ciufelli M, Kaleci S, Magliulo G, Presutti L, Re M. Clinical presentation and treatment outcomes of thyroglossal duct cysts: a systematic review. Int J Oral Maxillofac Surg. 2015;44(1):119–26.
26. Burkart CM, Richter GT, Rutter MJ, Myer CM 3rd. Update on endoscopic management of lingual thyroglossal duct cysts. Laryngoscope. 2009;119(10):2055–60.
27. Hirshoren N, Neuman T, Udassin R, Elidan J, Weinberger JM. The imperative of the Sistrunk operation: review of 160 thyroglossal tract remnant operations. Otolaryngol Head Neck Surg. 2009;140(3):338–42.
28. LiVolsi VA, Perzin KH, Savetsky L. Carcinoma arising in median ectopic thyroid (including thyroglossal duct tissue). Cancer. 1974;34(4):1303–15.
29. Heshmati HM, Fatourechi V, van Heerden JA, Hay ID, Goellner JR. Thyroglossal duct carcinoma: report of 12 cases. Mayo Clin Proc. 1997;72(4):315–9.
30. Plaza CP, López 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(5):745–52.
31. Forest VI, Murali R, Clark JR. Thyroglossal duct cyst carcinoma: case series. J Otolaryngol Head Neck Surg. 2011;40(2):151–6.
32. Zizic M, Faquin W, Stephen AE, Kamani D, Nehme R, Slough CM, Randolph GW. Upper neck papillary thyroid cancer (UPTC): 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. Laryngoscope. 2015. doi:10.1002/lary.25824.
33. Choi YM, Kim TY, Song DE, Hong SJ, Jang EK, Jeon MJ, Han JM, Kim WG, Shong YK, Kim WB. Papillary thyroid carcinoma arising from a thyroglossal duct cyst: a single institution experience. Endocr J. 2013;60(5):665–70.
34. Sullivan DP, Liberatore LA, April MM, Sassoon J, Ward RF. Epidermal inclusion cyst versus thyroglossal duct cyst: Sistrunk or not? Ann Otol Rhinol Laryngol. 2001;110:340–4.
35. Teissier N, Elmaleh-Bergès M, Ferkdadji L, François M, Van den Abbeele T. Cervical bronchogenic cysts: usual and unusual clinical presentations. Arch Otolaryngol Head Neck Surg. 2008;134(11):1165–9.
36. Kieran SM, Robson CD, Nosé V, Rahbar R. Foregut duplication cysts in the head and neck: presentation, diagnosis, and management. Arch Otolaryngol Head Neck Surg. 2010;136(8):778–82.
37. Johnston R, Wei JL, Maddalozzo J. Intra-thyroid thyroglossal duct cyst as a differential diagnosis of thyroid nodule. Int J Pediatr Otorhinolaryngol. 2003;67(9):1027–30.
38. Huang LD, Gao SQ, Dai RJ, Chen DD, He B, Shi HQ, Yang K, Shan YF. Intra-thyroid thyroglossal duct cyst: a case report and review of literature. Int J Clin Exp Pathol. 2015;8(6):7229–33.

39. Apel RL, Asa SL, Chalvardjian A, LiVolsi VA. Intrathyroidal lymphoepithelial cysts of probable branchial origin. Hum Pathol. 1994;25(11):1238–42.
40. Ko AB, Assaad A, Carty SE, Barnes EL, Schaitkin BM. Branchial cleftlike cysts of the thyroid. Am J Otolaryngol. 2006;27(2):146–8.
41. Carter E, Ulusarac O. Lymphoepithelial cysts of the thyroid gland. A case report and review of the literature. Arch Pathol Lab Med. 2003;127(4):e205–8.
42. Galluzzi F, Pignataro L, Gaini RM, Hartley B, Garavello W. Risk of recurrence in children operated for thyroglossal duct cysts: a systematic review. J Pediatr Surg. 2013;48(1):222–7.