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Thyroglossal Duct Cyst Carcinoma in The Pediatric Population: A Case Report and Literature Review

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ABSTRACT

Thyroglossal duct cyst carcinoma (TGDCa) is a rare cause of neck masses in children. Its pathophysiology stems from two main theories. One suggests that the primary carcinoma originated from ectopic thyroid tissue, whilst the other suggest it is a result of metastasis to the thyroglossal duct cyst from the thyroid gland. A case of a 12-year-old girl who presented with a suspicious neck mass and subsequently diagnosed with TGDCa is reported. A literature review was performed and we conclude that aggressive surgical management of a Sistrunk procedure, total thyroidectomy, bilateral tracheoesophageal groove clearance and neck dissection was justified for this child. We found that if the preoperative diagnosis is TGDCa, a more aggressive surgical approach tends to be taken. However, cases of recurrence remain low and a total thyroidectomy does entail a larger morbidity for the pediatric patient. Hence, we do not recommend an aggressive approach routinely. In this review, we extrapolated AJCC data for thyroid malignancies and staged the TGDCa reported in literature in the last 10 years. It showed us that pT3 tumors tend to have a more aggressive surgical course, whilst a pT1 TGDCa tend to receive a more conservative surgical approach.
Keywords: *Thyroglossal Duct Cyst Carcinoma, Papillary Thyroid Carcinoma, Pediatric Rare Diseases, Pediatric Surgery, Head and Neck, Endocrine, Endocrinology, Rare Thyroid Disease*

Introduction

Thyroglossal duct cyst carcinoma (TGDCa) is a rare disease in the pediatric population. This is in contrast to benign thyroglossal duct cysts which account for up to 75% of all midline neck swelling in children (Batazzi *et al*., 2019). TGDCa can present in a non-specific manner as a single painless anterior midline neck mass. Hence, diagnosis requires a high index of suspicion. Optimal management in this age

group can be highly debatable as there are no established guidelines for management of thyroglossal duct cyst carcinoma in the pediatric population (Thompson *et al*., 2017a). Even staging of thyroglossal duct cyst carcinoma is currently not advocated by the American Joint Committee of cancer (AJCC). In this case report, we present a case of a 12-year-old girl with papillary thyroid carcinoma in a thyroglossal duct cyst, review current literature and then justify our case of aggressive surgical management.

Case Description

A previously well 12-year-old girl was seen in the outpatient clinic for a midline neck lump of 3 years duration. Due to its painless and largely inconspicuous nature, the child and her parents were not worried until it began to grow rapidly in size in the prior few months. She had no complaints of constitutional symptoms such as fever, night sweats and weight loss. There were also no concerns in terms of airway and breathing. Family history was unremarkable for malignancy.

On examination, she conversed in full sentences with normal tone and pitch of voice for her age and gender. She had a normal body habitus with no apparent peripheral stigmata of thyroid disease. A firm 3cm X 3cm midline neck lump that was slightly skewed towards the right side was appreciated (Fig. 1). The lump was not fixed to the skin and there was no overlying skin changes. It moved with swallowing but not with tongue protrusion. There was no cervical lymphadenopathy felt. At this stage, her unusual presentation of a progressively enlarging neck lump led us to believe that this was unlikely due to a benign cause. Hence, the oncologists were involved at an early stage for preliminary investigations.

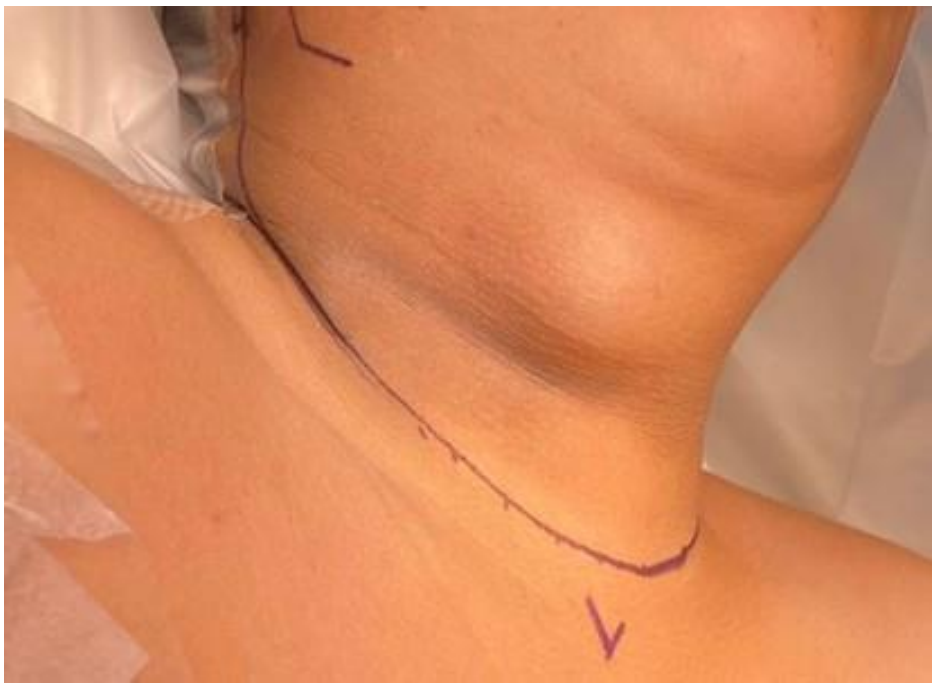

Figure 1: Clinical photograph of anterior neck lump that was more skewed to the right.

In terms of laboratory results, uric acid levels were slightly elevated at 357 umol/L, lactate dehydrogenase was normal at 188U/L. Hemoglobin was 13.3 g/dL, Total white cell count was 9.96x109/L and platelets was 311x109/L. She was euthyroid at time of visit with free serum thyroxine (fT4) of 12 pmol/L, serum thyroid stimulating hormone (TSH) at 1.19 mIU/L. Thyroglobulin (Tg) was elevated at 311 mIU/ml. Thyroid auto-antibodies were normal, with Tg antibodies (TgAB) <0.9 mIU/ml and TSH receptor antibodies (TRab) at 1.35 IU/L.

In terms of imaging, ultrasound (US) of the thyroid gland was performed and revealed a non-specific cystic lesion of 27cm X 2.4cm X 2cm superior to the thyroid gland. The cystic lesion was heterogenous in nature, with an internal solid component measuring 1.4cm X 1.3cm X 0.9cm (Fig. 2). Magnetic resonance imaging (MRI) of the neck was subsequently performed to further characterize the neck mass. It showed a complex lobulated solid cystic mass in the midline. Furthermore, the neck mass partially encased the hyoid bone, extending inferiorly along the strap muscles. However, it appeared separate from the thyroid gland and measured approximately 4.2cm X 3.6cm X 3.4 cm.

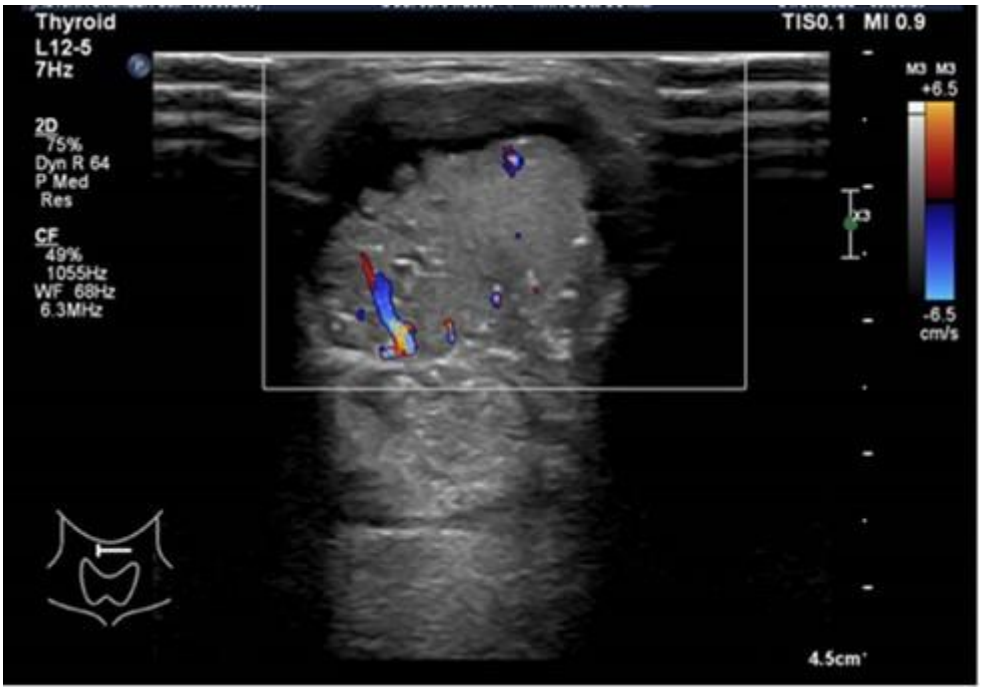

Figure 2: US of thyroid gland showing heterogeneous cystic lesion superior to the thyroid gland.

A computed tomography (CT) staging scan of the neck, chest and abdomen was performed and revealed no distant metastases. It demonstrated multiple enlarged lymph nodes at levels IB, IIA, III, IV, supraclavicular region of the right neck, and level IIA, III, IV, supraclavicular region of the left neck. The right cervical lymph nodes were concerning as they were seen compressing the right internal jugular vein (Fig. 3).

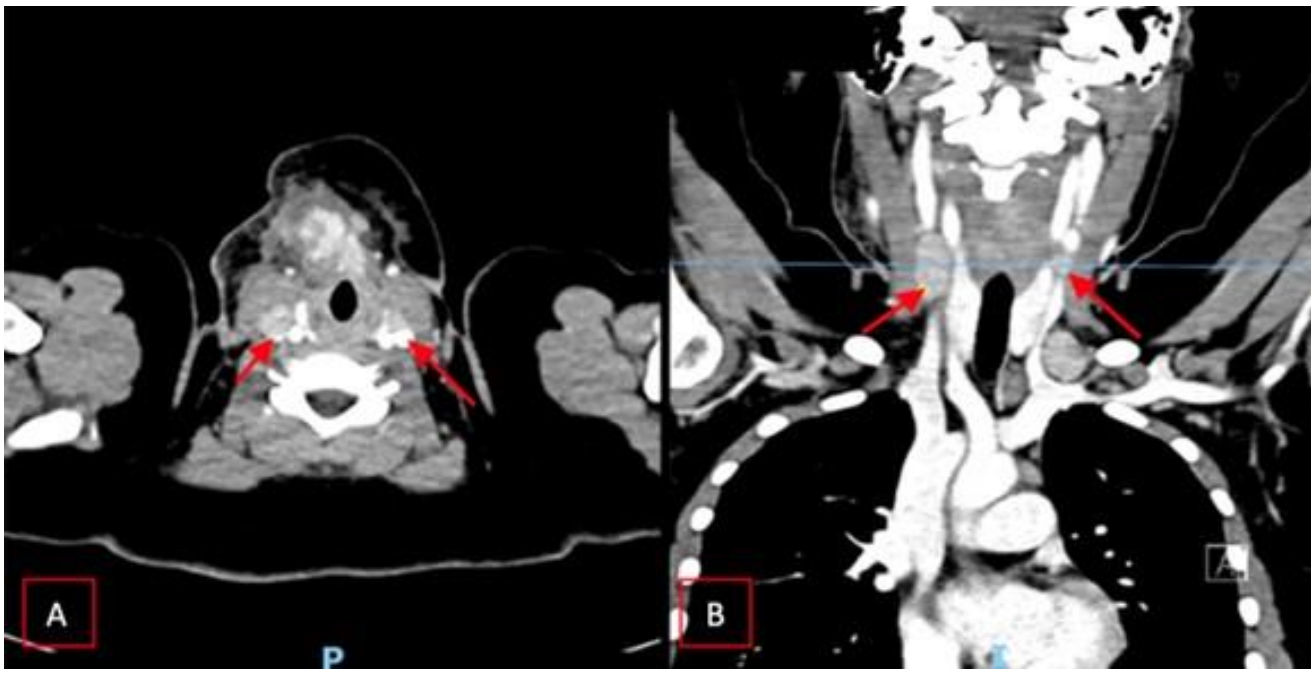

Figure 3: Computed tomography scans in axial (A) and sagittal (B) views demonstrate the complex lobulated solid cystic mass in the midline and the right internal jugular vein being compressed by cervical lymph node.

From imaging, we were convinced that there was malignant transformation within a thyroglossal duct cyst. Her case was discussed in a multidisciplinary tumor board meeting involving oncologists, pathologists, endocrinologists, and pediatric surgeons. Fine needle aspiration cytology of the neck mass revealed papillaroid clusters with enlarged nuclei, pale chromatin, intranuclear cytoplasmic pseudo-inclusions, nuclear grooves and a likely psammoma body (Fig. 4). The tru-cut biopsy allowed us to confirm a histopathological diagnosis of classical type papillary thyroid carcinoma within a thyroglossal duct cyst that was positive for Thyroid transcription factor 1(TTF 1) and Paired-box gene 8 (PAX 8) on immunohistochemical staining (Fig. 5).

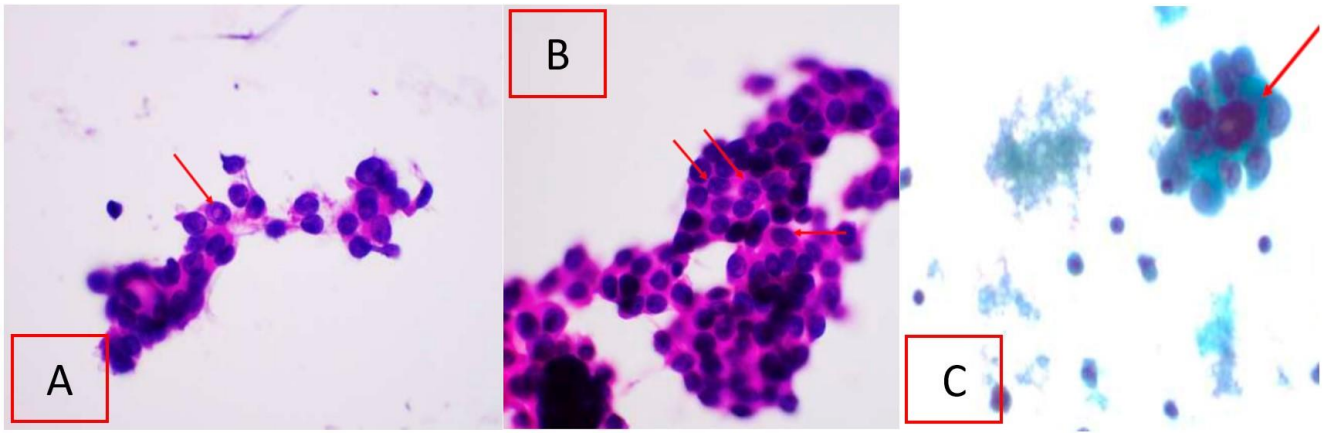

Figure 4: Fine needle aspiration cytology showing papillaroid clusters with enlarged nuclei, pale chromatin, intranuclear cytoplasmic pseudo-inclusions (A), nuclear grooves (B), psammoma body (C).

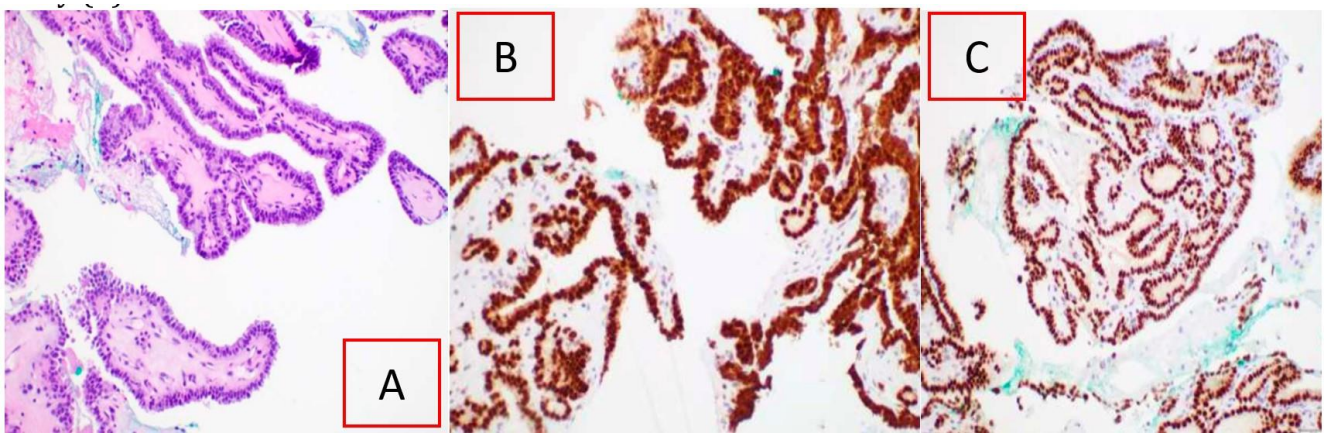

Figure 5: Tru-cut biopsy specimen (A) showing positivity for TTF1 (B) and Pax8 (C) immunohistochemical staining.

The patient underwent a Sistrunk procedure, total thyroidectomy, bilateral tracheoesophageal groove clearance and bilateral neck dissection (Fig. 6). Intraoperatively, the thyroglossal mass was found right to the midline, adhering to the hyoid bone and surrounding the strap muscles. On inspection, it did not invade the thyroid cartilage or thyroid membrane posteriorly. No tract to the foramen cecum was found. There were multiple enlarged cervical lymph nodes bilaterally in the neck, with one enlarged lymph node in the right carotid sheath, causing partial obstruction to the right internal jugular vein. Both upper and lower left parathyroid glands were excised and implanted to the right splenius capitis after histological confirmation by frozen section. The right parathyroid glands remained in situ. Crucial nerves such as the external branch of the superior laryngeal nerve, recurrent laryngeal nerves were identified bilaterally and preserved.

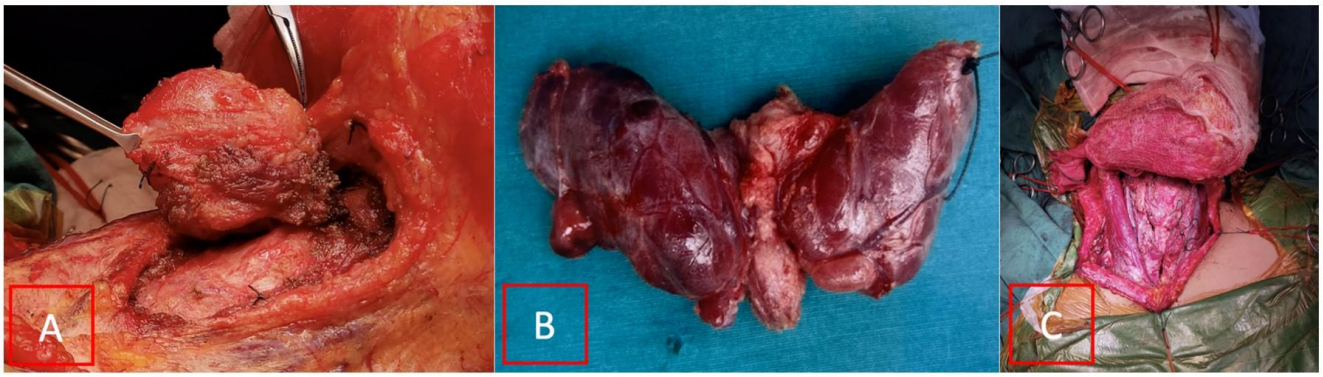

Figure 6: Sistrunk procedure (A), total thyroidectomy specimen (B) and bilateral tracheoesophageal groove clearance and bilateral neck dissection (C).

Post-operative histopathological evaluation of the thyroglossal mass showed a tumor measuring 3.8cm in its greatest dimensions with classical features of papillary thyroid carcinoma and clear resection margins. 9 out of 225 lymph nodes were positive of metastatic carcinoma, with 5 showing extra nodal extension. Extrapolation from AJCC 2010 criteria for thyroid gland malignancies, assigned our patient a stage of pT3/4N1bMx. Recovery was otherwise uneventful. She was extubated on postoperative day 1

and received intensive therapy from our allied health team including the speech and language therapists and physiotherapists on postoperative day 2. Clinically, there were no evidence of cranial nerve palsy. She was discharged on postoperative day 9.

Two months after surgery, 124I radioactive iodine (RAI) dosimetry scan was performed and the child went on to complete ablative 131I radioactive iodine treatment at a dose of 85mCi. With reference to Table 1, 2 months post-surgery, her serum Tg levels and TSH levels were suppressed at 54mIU/ml and 0.98mIU/L respectively. At 3 months post RAI treatment, her serum Tg levels was at 4.2mIU/ml and TSH was at 0.06 mIU/L. The down-trending Tg and TSH markers were reassuring.

Table 1: Analysis of Tg and TSH levels before surgery, 2 months post-surgery and 3 months post radioactive iodine treatment.

	Before surgery	2 months post-surgery/Start of RAI treatment	3 months post RAI treatment
Tg (mIU/ml)	311	54	4.2
TSH (mIU/L)	1.19	0.98	0.06

As a result of surgery, this patient developed hypocalcaemia secondary to hypoparathyroidism as well as hypothyroidism from total thyroidectomy. She was commenced on thyroid hormone supplementation, TSH suppression, calcium replacement and also close surveillance for cancer recurrence. The surveillance plan includes follow-up with the pediatric surgeons for wound healing, the oncologist for thyroid hormone and tumor marker screening and the nuclear medicine specialist for adjuvant RAI treatment. Currently, the patient is on monthly follow up with a pediatric oncologist, one year post surgery.

Discussion

During embryonic development, the thyroglossal duct connects the thyroid gland to the foramen cecum and the hyoid bone. The thyroglossal duct involutes and disappears gradually by the 10th week of development. If this does not occur, the thyroglossal duct remnants can persist anywhere along the pathway of thyroid descent. The epithelial cells that line the duct can then cause cystic dilatation and result in the formation of a thyroglossal duct cyst (TGDC). TGDCs make up 75% of midline neck masses in children. Occasionally, these TGDCs can harbor malignancy. Genetic mutations of interest that trigger malignant change include thyroid transcription factors TTF1, TTF2 and PAX 8 genes (Macchia, 2000). The normal function of these genes is for the development of thyroid follicular cells. This in turn forms the basis of two theories of origin of thyroglossal duct cyst carcinoma (TGDCa). They are, firstly, the de novo theory that suggests that the primary carcinoma arose from ectopic thyroid tissue in the thyroglossal duct cyst. In fact, in two-thirds of thyroglossal duct cysts, thyroid follicles are present histologically and only 1-2% have malignancy. The metastatic theory, on the other hand, postulates that a small occult primary

carcinoma of the thyroid gland has metastasized to the thyroglossal duct via lymphatics and hence resulted in a thyroglossal duct cyst carcinoma. (Tharmabala and Kanthan, 2013)

TGDCa in children is rare and management of TGDCa remains varied, with no consensus particularly on the best surgical and radiological management for these cases (Seow-En *et al*., 2015). For instance, there has been debate on whether a total thyroidectomy should be performed in addition to a Sistrunk procedure for pediatric patients with TGDCa is substantiated. The absence of a staging system for TGDCa makes it more challenging to determine clinical outcomes (Thompson *et al*., 2017a). The use of adjuvant radioactive iodine ablation has been reported but not evaluated. Hence, we suggest that a multidisciplinary team approach is crucial to decide on the most optimal management plan.

To evaluate our management plan in the context of evolving evidence, and understand how the management of thyroglossal duct cyst carcinomas has changed in the last 10 years, a literature search on PubMed was performed, using the search terms; (thyroglossal duct cyst carcinoma) OR (thyroglossal carcinoma). This yielded 543 articles from 1952-2022. We narrowed our search to articles published in the last 10 years from January 2012 to January 2022, and only included case reports of pediatric patients under the age of 21 (Table 2). This identified 20 patients in total over 10 years, aged 11-20 years.

Table 2: Literature review summary.

STUDY	AGE / SEX	TUMOR SIZE (CM)	TNM STAGING	PRE-OP INVESTIGATIONS	PRE-OP DIAGNOSIS	POST OP HISTOPATHOLOGY	SURGICAL MANAGMENT	ADJUVANT THERAPY	OUTCOME		
									RECURRENCE	FOLLOW UP DURATION	FOLLOW UP METHOD
Pandey, *et al*. (2021)	11/F	5.5 by 4 by 5	pT3aN1Mx	US, FNAC, CT	TGDC	PTC. LVI	SP	N	N	2y	US
Boyanov, *et al*. (2020)	17/F	3 by 2	pT2NxMx	US	NA	PTC, invasion of cyst wall	Excision of neck mass, with preservation of neck muscles and hyoid bone	N	N	10m	US
Liaw, *et al*. (2019)	15/ M	NA	NA	US, CT	NA	PTC, metastasis or direct extension from thyroid primary cannot be excluded	SP followed by TT 1 month later	ATI	N	NA	NA
El Korbi, *et al*. (2019)	14/F	1	pT1NxMx	US, CT	NA	PTC, invasion of adjacent muscles	SP followed by TT	ATI and LTS	N	1y	RAI total body scan, serum Tg
Thompson, *et al*., (2017b)	16/ M	0.6	pT1NxMx	XR. CT	TGDC	PTC, invasion of cyst wall and adjacent muscles	SP followed by TT 8 months later	N	N	11m	NA
	12/ M	0.1	pT1NxMx	CT, MRI	NA	PTC, invasion of cyst wall and adjacent muscles	SP	N	N	22m	NA
Cheon, *et al*. (2016)	17/F	2	pT1bNxMx	CT	TGDC	PTC	SP	NA	NA	NA	NA
Al-Hadidi, *et al*. (2016)	12/F	NA	NA	US, CT	NA	PTC, LVI	SP, WLE, regional lymphadenectomy	N	N	6m	Imaging, TFTs
Hassan, *et al*. (2016)	17/F	1 by 0.8	pT1aNxMx	US	Infected TGDC	PTC	SP	N	N	1y	US

Tahir, *et al*. (2015)	10/F	9	pT1N0Mx	US	TGDC	PTC, LVI	SP followed by TT	ATI and LTS	N	1y	Clinical exam
Shankar, *et al*. (2015)	17/F	4 by 3 by 2	pT3NxMx	US	TGDC	PTC, invasion of adjacent soft tissue	SP	NA	NA		
Seow-En, *et al*. (2015b)	15/ M	8.5	pT3N1Mx	XR, CT, MRI, US FNAC	Lymphangioma vs rhabdomyosarcoma	PTC, LVI	SP with enbloc resection of the mass, TT, bilateral radical neck dissection	ATI and LTS	N	NA	Clinical exam and serum Tg
Pezzolla, *et al*. (2015)	20/F	NA	pT1N0M0	US FNAB	TGDCa, PTC	PTC, invasion of cyst wall	Removal of sub hyoid mass and hyoid bone and TT	ATI and LTS	N	1y	serum Tg, US
Diaconescu, *et al*. (2015)	14/F	NA	NA	US FNAB	TGDC	PTC, invasion of cyst wall	SP	N	N	NA	NA
Vassilatou, *et al*. (2014)	19/ M	1.8	pT1bNxMx	CT, MRI, US FNAC	TGDCa, PTC	PTC	SP and TT	ATI (70mCI I 131) and LTS	N	4y	US, anti-thyroglobulin antibodies, serum Tg
Proia, *et al*. (2014)	20/F	NA	NA	US	TGDC	PTC	Modified SP followed by TT	ATI (2 Rounds)	N	1y	NA
Pfeiffer, *et al*. (2014)	15/F	2.3 by 2 by 2	pT2N0Mx	CT, I 131 scan, US FNAC	TGDCa, PTC	PTC, invasion of skeletal muscle	SP with WLE	NA	NA	NA	NA
Chrisoulidou, *et al*. (2013)	18/ M	0.8	pT1NxMx	US	NA	PTC	Surgical resection of neck mass	NA	Y	10y	US
	18/F	NA	NA	US	NA	PTC, invasion of adjacent fat tissue	Surgical resection of neck mass	NA	Y	NA	US
Dan, *et al*. (2012)	18/F	4	pT2N0M0	US, CT	TGDC	PTC, incomplete invasion of cyst wall	SP followed by TT 2 months later	LTS	N	NA	serum Tg, RAI scan

Key: FNAC: fine needle aspiration cytology, FNAB: fine needle aspiration biopsy, PTC: papillary thyroid carcinoma, LVI: lymphovascular invasion, SP: Sistrunk procedure, TT: Total thyroidectomy, WLE: wide local excision, ATI: ablative radioactive iodine, LTS: Levothyroxine suppression. TFTs: Thyroid function tests, Tg: thyroglobulin, RAI: radioactive iodine, US: Ultrasound, CT: computed tomography, Y: Yes, N: No, NA: Not applicable

Firstly, we discuss the need for a total thyroidectomy in pediatric patients with TGDCa. In the literature, 6 patients (30%) had staged operations (i.e., a Sistrunk operation followed by a total thyroidectomy) as post-operative histopathology showed microscopic invasion of the thyroid gland and a total thyroidectomy would facilitate the use of adjuvant radioactive iodine. Only 3 patients (15%) had a concurrent total thyroidectomy. The most common reason for doing a concurrent total thyroidectomy was due to abnormal thyroid findings either on physical exam or on gross inspection intraoperatively. Out of all the cases that had total thyroidectomies, either as a concurrent or staged operation, only Pezzolla, *et al*. (2015) reported positive findings of an incidental synchronous thyroid adenocarcinoma. Chrisoulidou, *et al*. (2013) performed a total thyroidectomy subsequently as a result of TGDCa recurrence. Thompson, *et al*. (2017b) reported retrospective identification of ectopic thyroid tissue in a TGDC specimen that was histologically proven to be papillary thyroid carcinoma, though a total thyroidectomy was not performed at the time. Thus, considering the significant morbidity and need for lifelong hormone replacement as a result of total thyroidectomy, the emerging evidence does weigh against routine concurrent total thyroidectomy for children with TGDCa.

Only 2 patients (10%), discussed in Chrisoulidou, *et al*. (2013), with TGDCa recurrence underwent subsequent total thyroidectomies. The first case was an 18-year-old boy who underwent surgical resection of a neck mass that demonstrated papillary type TGDCa. 10 years after surgery, he noted rapid thyroid swelling and subsequently underwent a concurrent Sistrunk procedure and total thyroidectomy. The second case was an 18-year-old girl who initially had a suspicious neck mass removed that demonstrated to be TGDCa of papillary thyroid origin; with invasion of adjacent fat tissue. She later presented again with bilateral cervical lymphadenopathy. Hence total thyroidectomy and modified bilateral lymph node dissection was offered and performed. These cases of recurrence remain rare and are likely associated with a missed diagnosis of TGDCa prior to the primary surgery and incomplete resection at primary surgery.

Decisions for total thyroidectomy could be influenced by the initial differential diagnosis. 9 patients (45%) had a pre-operative diagnosis of TGDC, but a post-operative diagnosis of TGDCa. Of these 9 patients, only 4 patients (44.4%) underwent a total thyroidectomy in a second operation. In contrast, out of 3 patients who had a pre-operative diagnosis of TGDCa, 2 patients (66.7%) underwent a concurrent total thyroidectomy. Hence, if a diagnosis of TGDCa is made preoperatively, the child seems more likely to undergo a more aggressive operation with concurrent total thyroidectomy. Notably, out of 12 patients (60%) who did not undergo total thyroidectomy, 1 patient was offered to do so, but did not proceed due to parental refusal. Therefore, shared decision making regarding surgical choices should be made with parents, ideally together with the young patient, as much as the latter may be able to understand.

Cancer staging such as the tumor, nodal, metastases (TNM) staging system is an internationally accepted system to characterize the extent of disease. It allows clinicians to estimate cancer survival and might have a role in providing treatment recommendations for a rare disease such as TGDCa. However, staging for thyroglossal duct cyst carcinoma is not advocated or included in the 8th edition of the AJCC staging manual (Thompson *et al*., 2017a). Thompson, advocated for staging TGDCa using extrapolated data from the AJCC 2010 criteria for thyroid gland malignancies, and that this will help to risk stratify patients and guide treatment decisions in view of the various therapeutic options and dilemmas surrounding this rare disease. In our case, using extrapolated data from AJCC 2010 criteria for thyroid gland malignancies, we staged our patient as pT3/4 N1b Mx. In the literature reviewed, only one other author, Boyanov, *et al*. (2020) performed tumor staging based on this extrapolated data. This shows that for most authors, surgical management was mostly dictated based on clinical experience, and less so, TNM staging.

In an effort to objectively compare management differences, we staged the reviewed cases by extrapolating the AJCC TNM staging data, where exact histological size of the tumors was available. There were 2 patients (10%) with pT3 TGDCa, and no patients with pT4 TGDCa. In the pT3 cases, Sistrunk procedure was performed, and total thyroidectomy was advocated. In Pandey *et al*., 2021, the patient did not undergo total thyroidectomy as parents refused a second operation. Shankar *et al*., 2015 were convinced about the patient needing a subsequent total thyroidectomy, radioactive iodine ablation and thyroxine suppression. However, this patient only reportedly underwent a Sistrunk procedure and it is unclear if the patient underwent further treatment. In all restaged pT3 cases, there were either no mention or no reported cancer recurrences. Hence, for cases staged by extrapolated AJCC data as pT3, patients tend to be offered both Sistrunk procedure and total thyroidectomy. In contrast, there were more varied approaches with pT1 tumors, with 3 patients (15%) that had Sistrunk procedure alone; 3 patients (15%) with staged Sistrunk procedure and total thyroidectomy; 2 patients (10%) with concurrent total thyroidectomy in the same setting and 1 patient (5%) who underwent surgical resection of neck mass. Chrisoudilou's case of a pT1 tumor that recurred only after 10 years with subsequent thyroid involvement further supports that the influence of staging, (using extrapolated data to begin with), on treatment is complex and not necessarily helpful especially for smaller pT1 tumors.

In terms of follow up, most authors remain vague with regards to follow up duration and methods. For instance, Pandey *et al*., 2021 reported that lymphovascular invasion was noted post operatively after a Sistrunk procedure and that parents declined total thyroidectomy and bilateral lymph node dissection. However, the authors wrote that after 2 years of follow up, the patient's repeat US of neck did not show any abnormality in the thyroid or lymphadenopathy.

Out of 11 patients (55%) who underwent total thyroidectomies (including the 2 cases that performed total thyroidectomies as a result of cancer recurrence), only 5 patients (45.5%) went on to have adjuvant radioactive iodine and 2 patients (18.1%) used radioactive iodine scans to monitor recurrence. There were insufficient details available to make conclusions regarding the utility of adjuvant radioactive iodine and the benefit of using radioactive iodine scans as a modality for follow up in TGDCa patients who underwent total thyroidectomies.

Conclusion

Thyroglossal duct cyst carcinomas are rare cancers in the pediatric population with intricate dilemmas about management options since the first reported case by (Brentano H, 1911). This paper examined the trend of management of TGDCa in the last 10 years and found that a more conservative approach is favored, with most cases undergoing only a single Sistrunk procedure for TGDCa. Whilst there are no formal staging guidelines for TGDCa, we suggest extrapolating AJCC staging data and to consider a more aggressive approach with total thyroidectomy for tumors staged pT3 and above. For pT1 tumors, shared informed decision making involving the pediatric patient, their parents, and a multidisciplinary team involving surgeons, radiologists and oncologists remains an imminent part of the decision-making process for a personalized approach to each case.

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