

## Original Article

# Papillary thyroglossal duct carcinoma: report of nine cases and review of literature

Ying-Ying Zhu<sup>1\*</sup>, Cheng-Gang Wang<sup>2\*</sup>, Wu-Yi Li<sup>1</sup>, Zhi-Qiang Gao<sup>1</sup>, Xing-Ming Chen<sup>1</sup>

<sup>1</sup>Department of Otolaryngology-Head and Neck Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences & Peking Union Medical College, Beijing, China; <sup>2</sup>Department of Otolaryngology-Head and Neck Surgery, Qujing Second People's Hospital of Yunnan Province, Yunnan, China. \*Equal contributors.

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**Abstract:** Background: Thyroglossal duct carcinoma (TDCa) is a rare malignancy. Thus, we summarized the clinical characteristics of TDCa to explore the strategy of diagnosis, surgical treatment, and prognosis of patients with papillary TDCa. Case presentation: A retrospective study was conducted about the medical records of cases with TDCa. General information including the diagnosis, surgical treatment, and prognosis of patients were obtained and analyzed. A total of 480 patients with thyroglossal duct anomalies were treated in our hospital during 1981 to 2011. Nine patients were identified as papillary TDCa and Sistrunk procedure was performed in these patients. Total thyroidectomy and selected neck dissection were performed in 3 TDCa patients with additional multiple thyroid nodules at presentation, of whom one was diagnosed as papillary thyroid carcinoma and another was found to have lymph node metastases. With a median follow-up of 119 months, all patients were alive with no recurrences or metastases. Conclusions: Although the incidence of TDCa is very low, early diagnosis and surgical operation of TDCa should be performed as soon as possible. Besides, the pathological examination is the only way to confirm the diagnosis, and surgery, especially Sistrunk's procedure, is the prior choice of the treatments. In the presence of thyroid lesions or cervical lymphadenopathy, total thyroidectomy and neck dissection should be considered.

**Keywords:** Papillary thyroglossal duct carcinoma, diagnosis, treatment, head and neck neoplasms

## Introduction

Thyroglossal duct cysts, as the most common congenital neck mass, can form any-where along the thyroid's route of migration, but are most commonly found inferior to the body of the hyoid bone [1, 2]. Thyroglossal duct carcinoma (TDCa) is a malignant tumor that occurs within the thyroglossal duct remnant [3]. Besides, TDCa is extremely rare, occurring in approximately 1% of thyroglossal duct cyst cases, with the most common type being papillary carcinoma [4, 5]. Therefore, there have been few large series of TDCa reported in the previous literature.

It is efficient to make a correct preoperative diagnosis by the careful clinical history and physical examination in most situations [6]. However, some controversial issues regarding origin, diagnosis, and treatment still remain unclear. In recent years, the number of the

studies about papillary TDCa was relatively small, resulting in no enough statistical sample size to clarify the clinical characteristics of this disease. Hence, the statistical analysis of these data could be more accurate and objective evaluation to provide more evidence, which was very important for clinicians. Hence, the aim of this study was to improve the understanding of the clinical features of TDCa by analyzing the medical records of patients who were diagnosed with TDCa in our hospital. In addition, we studied previous relevant researches about TDCa and made a literature of review.

## Case presentation

As TDCa is a rare disease, no randomized controlled studies were retrieved. Relevant keywords were retrieved in the databases PubMed, covering all the papers published until May 1st, 2017, and a total of 50 studies about the cases of papillary TDCa were detected (31 men, and

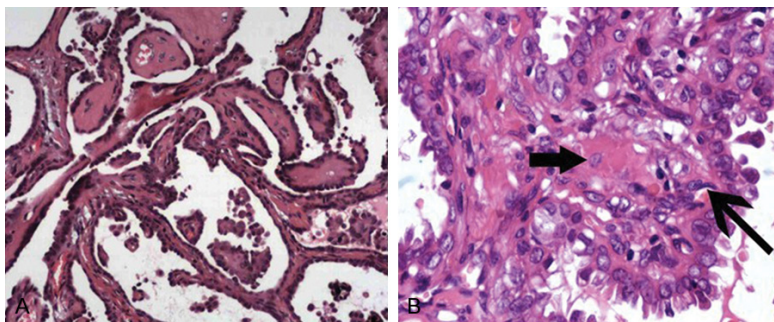
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**Table 1.** Characteristics of individual studies included in the literature review

Author	Year	Country	No of cases	Median/ Mean age	Sex (Male/Female)	Type of operation	Follow-up (months)
Higgins	2017	America	1	40	M	-	-
Penna	2017	Brazil	1	36	F	TT	9
Srivanitchapoom	2017	Thailand	1	38	F	TT	12
Yoo	2016	America	1	41	F	-	6
Hassan	2016	India	1	17	F	SO	>12
Medina	2016	Mexico	1	46	F	TT	-
Baglam	2015	Turkey	1	39	F	SO+TT	60
Sobri	2015	Indonesia	1	57	M	SO+TT	-
Proia	2014	Italy	1	20	F	TT	>12
Maleki	2014	Iran	1	22	F	SO	24
Pfeiffer	2014	America	1	15	F	SO	-
Vassilatou	2014	Greece	1	19	M	SO+TT	12
Choi	2013	Korea	10	52 (25-72)	2 M, 8 F	4 SO, 6 SO+TT	58.1 (4-165)
Yang	2013	Korea	1	28	F	SO	36
Aculate	2013	United Kingdom	1	21	F	SO+TT	-
Senthilkumar	2013	India	1	52	M	SO	-
Tharmabala	2013	Canada	1	32	F	SO	12
Hakeem	2013	India	1	40	F	SO+TT	36
Pietruszewska	2012	Poland	1	64	M	SO	192
Gomi	2011	Japan	1	11	F	SO	10
Balalaa	2011	America	1	31	M	SO+TT	-
Torcivia	2010	France	2	54 (47-60)	1 M, 1 F	TT	48
Ogawa	2010	Japan	1	61	F	SO+TT	-
Albayrak	2010	Turkey	1	39	-	TT	6
Park	2010	Korea	1	46	F	SO+TT	6
Mesolella	2010	Italy	1	27	M	SO	-
Hartl	2009	France	18	41.5	5 M, 13 F	2 SO, 15 TT, 1 isthmusectomy	144 (12-264)
Smiti	2009	India	1	25	F	SO	-
Murali	2009	India	1	23	M	TT	6
Cherian	2009	India	1	58	F	SO+TT	6
Heemskerk	2009	Netherlands	1	30	M	SO	-
Aghaghazvini	2009	Iran	1	44	F	-	12
Ishay	2008	Israel	1	61	M	SO	24
Kandogan	2008	Turkey	1	44	F	SO	24
Canani	2008	Italy	1	35	F	SO	24
Aggarwal	2007	India	1	28	F	SO	-
Ali	2007	Libya	1	40	M	SO+TT	-
Banerjee	2007	India	2	28 (26-30)	2 F	SO	>12
Luna-Ortiz	2004	Mexico	5	49	3 M, 2 F	SO	-
Naghavi	2003	Iran	1	28	M	SO+TT	-
Ramalingam	2003	India	1	22	F	SO	12
Öztürk	2003	Turkey	1	11	M	SO	6
Datar	2000	India	1	35	M	SO	-
Martins	1999	Brazil	1	21	F	SO	24
Buchi.o	1999	America	1	17	-	-	-
Thakar	1999	India	1	29	M	SO	12
Kojima	1995	Japan	1	34	F	-	-
Kwan	1995	Canada	1	38	M	TT	36
Thornton	1989	Northern Ireland	1	65	M	-	-
Mcnicoll	1987	America	1	9	M	TT	18

SO: Sistrunk operation; TT: Total thyroidectomy; M: Male; F: Female.

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**Figure 1.** Histopathology results of papillary TDCa. A. Microscopic examination shows typically slender arborizing papillae formations (H&E, 60 ×). B. Ground glass nuclear (short arrow) and nuclear groove (long arrow; H&E, 400 ×).

51 women). Demographic and clinical characteristics of patients were shown in **Table 1**. Among these, Sistrunk operation was performed in 24 studies. In addition, the patients in 10 studies underwent total thyroidectomy, and Sistrunk operation with total thyroidectomy was performed in 12 studies. Moreover, no recurrences or metastases found in all patients. The median follow-up period was 46 months (arrange from 3 months to 700 months).

Patients diagnosed with TDCa and received treatment in our hospital between 1981 and 2011 were reviewed retrospectively. Histopathology results of papillary TDCa were showed in **Figure 1**. In histological examination, we observed slender arborizing papillae formations, ground glass nuclear and nuclear groove in the patients with TDCa. In addition, the expression levels of thyroid protein, thyroid transcription factor-1 and Galectin-3 were observed in these papillary TDCa patients. The results of immunohistochemical staining of papillary TDCa tissues were showed in **Figure 2**. All the available information were collected and the extracted elements from the medical records included: age, gender, clinical presentation, tumor site, preoperative thyroid lesions, preoperative lymph nodes, postoperative thyroid lesions, postoperative lymph node metastasis, initial treatment, follow-up treatment, follow-up and vital status. In addition, the follow-up of all patients ended on December 30, 2016. All histological samples were reconfirmed by two senior pathologists.

In this study, nine patients were diagnosed with TDCa during 1981 to 2011 (4 men, and 5 women), and a total of 480 patients with thyro-

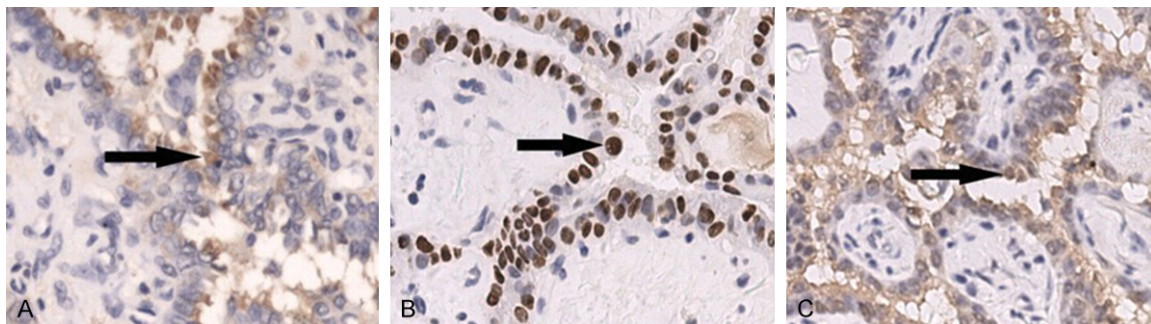
glossal duct anomalies were found in the same period. TDCa accounted for 1.875% of the thyroglossal duct anomalies. Demographic and clinical characteristics of nine patients with TDCa were shown in **Table 2**. The median age of 9 cases ranged from 27 to 64 years (an average age of  $44.6 \pm 11.2$  years). The chief complaint at presentation was a painless mass in the anterior of the neck, and thyroid glands

were found in the anterior of the trachea. Sistrunk procedure was performed in all 9 patients. Among them, three patients presented with additional multiple thyroid nodules received total thyroidectomy and selected neck dissection, of whom one was diagnosed as papillary thyroid carcinoma and another was found to have lymph node metastases. Moreover, all malignant lesions were pathological diagnosed as papillary TDCa, and no surgery-associated complications occurred in all patients. After the operation, all patients with total thyroidectomy received suppressive doses of thyroxine. Besides, radioactive iodine therapy was carried out in the patients with postoperative lymph node metastases. The median follow-up period was 65 months to 218 months (average  $126.8 \pm 49.9$  months). What's more, all nine patients were alive with no recurrences or metastases.

### Typical case

A 35-year-old male presented with a 2-year history of the mid-line neck mass. The patient was completely asymptomatic and had no significant past medical, surgical history and family history of thyroid disease or history of head and neck irradiation. Physical examination revealed a moveable, smooth and cystic swelling mass of 4.0 cm × 3.0 cm × 3.0 cm in the anterior of the neck, and multiple thyroid nodules were found. In addition, bilateral neck lymph nodes were also palpated and there was no associated cervical lymphadenopathy. Before hospitalization, hyperthyroidism has been diagnosed 11 years ago. Subsequently, the patient developed hypothyroidism after treatment with iodine-131 ( $^{131}$ ). Over the past several years,

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**Figure 2.** Immunohistochemical staining of papillary TDCa tissues. The brown granule staining indicates a positive reaction for each protein (arrows; 400 ×). A. Thyroid protein; B. Thyroid transcription factor-1; C. Galectin-3.

this patient was always taking oral levothyroxine and thyroid function maintained within the normal range.

In the present, the chest radiography showed no abnormal findings. Laboratory examinations including thyroid hormone were normal, indicating that the patient's thyroid function was normal. A computed tomography (CT) scan of the neck found a heterogeneous lesion at the level of the hyoid bone, with calcification and enhancement. In addition, the thyroid radionuclide scan revealed no uptake function in this neck mass. Neck ultrasound showed multiple thyroid nodules, a well-demarcated, heterogeneous mass in the anterior neck, and lymphadenopathies in the bilateral neck with punctate calcification. Meanwhile, the biopsy of the anterior neck mass indicated a papillary carcinoma of the thyroglossal duct.

Subsequently, the patient underwent Sistrunk's procedure (resection of the cyst and removal of the body of hyoid bone), as well as total thyroidectomy with bilateral neck dissection. The result of pathology examination confirmed the diagnosis of a papillary TDCa and cervical metastases. What's more, the histopathological examination of the excised thyroid gland revealed nodular goiter, with no evidence of cancer formation. Thyroid suppression treatment and radioactive iodine therapy with  $^{131}\text{I}$  ablation were administered after the surgical procedures. There was no evidence of recurrence or metastasis after 76 months' follow-up until now, and the patients did not find any discomfort and radiographic abnormalities.

### Discussion

Thyroglossal duct cysts (TDCs) represents the most common congenital neck mass which

often occur in pediatric patients, and is found in approximately 7% of the population [57]. During the sixth week of embryogenesis, the thyroglossal duct descends from the foramen cecum of the tongue into its location in the anterior neck. It reaches its final position in front of the upper trachea, and subsequently is obliterated. If the duct fails to atrophy, it may give rise to thyroglossal duct cysts [58]. About 1% of thyroglossal duct cysts eventually develop neoplastic changes, and carcinomas arising from these remnants are rare [59]. TDCs can present at any age and patients typically present with a slow growing painless mass that is mid-line and moves with swallowing and tongue protrusion [60]. In 480 cases of thyroglossal duct anomalies, we found nine cases of TDCa with an incidence of 1.875%. Among them, the most common histological pattern found (80%) is papillary TDCs. Other histological types include mixed follicular and papillary carcinoma, squamous cell carcinoma, follicular carcinoma, anaplastic carcinoma, and Hürthle cell carcinoma [61]. In our study, all nine cases in our hospital were papillary TDCs. Thus, we summarized the clinical characteristics of papillary TDCa to explore the strategy of diagnosis, treatment, and prognosis of these patients.

Due to the small number of reported cases, it is difficult to draw conclusions regarding management of this rare disease, indicating that this remains an area of controversy. Since it has implications in disease management, the anatomical origin of TDCa is important for the clinicians. One proposal proposes that the TDCa develops *de novo* from native thyroid tissue found within the walls of the thyroglossal duct cyst [62]. Another proposal is that TDCa is metastatic from that an occult primary thyroid gland and the thyroglossal duct cyst could act

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**Table 2.** Clinical features of nine cases of TDCa

Case number	Age (years)	Sex	Mass Size (cm)	Pre-op thyroid lesions	Pre-op lymph nodes	Initial treatment	Post-op thyroid lesions	Post-op lymph node metastasis	Follow-up treatment	Follow-up (months)/vital status
1	35	M	4.0 × 3.0 × 3.0	P	P	SP, T, and ND	NG	POS	Suppression, <sup>131</sup> I	79/NED
2	64	M	4.0 × 3.0 × 2.5	P	A	SP, T, and ND	PTC	NEG	Suppression	62/NED
3	45	F	2.5 × 2.0 × 2.0	P	A	SP, T, and ND	NG	NEG	Suppression	135/NED
4	37	F	2.0 × 1.0 × 1.0	A	A	SP	NP	NP	None	147/NED
5	46	M	3.0 × 3.0 × 2.5	A	A	SP	NP	NP	None	183/NED
6	56	F	4.5 × 2.5 × 2.0	A	A	SP	NP	NP	None	121/NED
7	27	F	1.5 × 2.0 × 1.0	A	A	SP	NP	NP	None	101/NED
8	42	M	2.0 × 2.0 × 1.5	A	A	SP	NP	NP	None	218/NED
9	50	F	3.0 × 2.0 × 2.0	A	A	SP	NP	NP	None	102/NED

Pre-op-preoperative; Post-op-postoperative; M-male; F-female; P-present; A-absent; SP-Sistrunk procedure; T-thyroidectomy; ND-neck dissection; NG-nodular goiter; PTC-papillary thyroid carcinoma; NP-not performed; POS-positive; NEG-negative; <sup>131</sup>I-iodine 131; NED-no evidence of disease.



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as a natural conduit for the spread of thyroid carcinoma [63]. Most TGDCs contain normal thyroid tissue and have been papillary or follicular, which provides support to the former theory. Due to the high incidence of concurrent thyroid carcinoma in patients with TGDCs, the latter theory is more widely supported than the former, and suggests that the fibrous tract may itself be a route of spread [64]. In addition, since the thyroglossal duct is normally an embryological structure, TDCa and thyroid carcinoma may be disseminated concurrently [65, 66]. In the present study, the total thyroidectomy was performed in three patients, and additional thyroid carcinoma was found in one case of TDCa.

Because the clinical profile of TDCa is similar to thyroglossal duct cyst, the differential diagnosis is necessary [67]. The most common symptom of TDCa is the presence of an anterior neck mass. Moreover, cancer onset time ranges from several months to years. Combined relevant researchers found that 70% of TDCa cases had no symptoms, the mean age of presentation was 40-60 years, and the gender ratio of female/male was 1.5:1 [62, 68]. Final diagnosis often relied on the histopathologic analysis of the specimen. Therefore, we believed that malignancy should be suspected, if the cyst was hard, fixed, irregular, or suddenly expands with palpable neck lymph nodes.

Regarding the use of CT scans and magnetic resonance imaging (MRI) in TDCa patients, one study showed that the presence of a midline cystic lesion with a dense or enhancing mural nodule, or calcification, should arouse the suspicion of carcinoma in a thyroglossal duct cyst patient [69]. Another study found that 3-dimensional CT made it possible to clarify the anatomical relationship between the tumor tissue and adjacent tissue, thereby allowing for an accurate preoperative diagnosis [70]. In the TDCa patients, a heterogeneous cystic lesion within areas of inhomogeneous enhancement was observed on CT scans. When some imaging features were observed, such as a mural lesion in the cyst with irregular margins or calcifications, sometimes with enhancement, we reasoned that malignancy should be considered in the patient with a thyroglossal duct cyst. In addition, the neck ultrasound revealed lymphadenopathies in bilateral neck, and some

with punctuate calcification in one patient. Therefore, these might be an indication of lymph node metastases. We suggested that multiple diagnostic tools were required for the accurate diagnosis and differential diagnosis from the other malignancies. Currently, preoperative fine needle aspiration biopsy (FNAB) is considered as the main diagnostic tool for TDCa, with a true-positive rate of 60% [71]. Besides, this technique was reported to be safe and reliable for the diagnosis of solid tumors other than cystic lesions [72]. No matter what, the pathological examination was the only way to confirm the diagnosis of TDCa.

The most appropriate surgical treatment for papillary TDCa is Sistrunk's operation. This procedure was first proposed in 1920, and required simultaneous excision of the mass as well as the entire thyroglossal tract and a portion of the hyoid bone nearby [73]. This procedure has shown a high cure rate of 95% for papillary TDCa, and often provided that there was no clinical or imaging suspicion of thyroid lesion or cervical adenopathies [74]. Some studies suggested that thyroidectomy could be considered for patients with multiple thyroid nodules [75]. However, in the current series, three patients had additional multiple thyroid nodules and underwent Sistrunk's procedure and total thyroidectomy, and another one case was diagnosed with papillary TDCa. Since all patients were free of disease up to now in this study, partial or total thyroidectomy might not be necessary for those with no thyroid abnormalities at presentation. In addition, when the patients had cervical lymphadenopathy, modified or radical neck dissection is necessary, and concurrent total thyroidectomy should also be considered. In this study, all patients in this cohort were alive with no recurrences or metastases. The more common papillary TDCa has an excellent prognosis with complete excision being curative [76]. Long-term follow up and a multidisciplinary approach are essential in all cases [77]. Additional high-quality and multicenter studies are required to better delineate management guidelines for this rare and controversial malignancy.

### Conclusions

TDCa is a rare malignant tumor that occurs within the thyroglossal duct remnant. It is

important for TDCa in the term of the differential diagnostic procedure, and the pathological examination is the only way to confirm the diagnosis of TDCa. Early diagnosis and surgical operation should be performed as soon as possible. Therefore, Sistrunk's operation of thyroglossal excision is reliable and the first-line choice for treatment. Moreover, TDCa cases with thyroid lesions or cervical lymphadenopathies, thyroidectomy and neck dissection should be considered. Further researches and clinical trials with larger sample sizes are needed to confirm our findings in subsequent articles.

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Written informed consent was obtained from the patients for the publication of this case report and any accompanying images.

### Disclosure of conflict of interest

None.

**Address correspondence to:** Xing-Ming Chen, Department of Otolaryngology-Head and Neck Surgery, Peking Union Medical College Hospital, Chinese Academy of Medical Sciences & Peking Union Medical College, 1 Shuaifuyuan, Wangfujing, Beijing 100730, China. Tel: +86-10-69156328; Fax: +86-10-69156311; E-mail: xiingming.chen@hotmail.com

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