

Thyroglossal Duct Cyst Revisited: A 13-Year Clinical Audit and Review of Literature

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Introduction

Thyroglossal duct cysts (TDCs) are the most common congenital anomaly of the neck in childhood, representing more than 70% of congenital neck anomaly [1]. The cyst usually is a painless midline mass close to hyoid bone and slightly mobile. In most cases, the patient just present with an asymptomatic soft mass. The cyst may also develop anywhere along the path of a persistent thyroglossal duct: 60% are located between the hyoid bone and thyroid gland, 24% are suprahyoid, 13% are suprasternal and 1% is intralingual [2]. Movement of cyst with protrusion of tongue with stabilization of the mandible is a reliable diagnostic sign. On occasional basis, some patients present with the complications of infected cyst or fistulae. Rarely, airway obstruction and dyspnea may occur especially if the mass is located at the base of tongue [2].

A meticulous clinical history and physical examination are usually sufficient to make a correct preoperative diagnosis; furthermore a preoperative imaging is an important step to confirm the diagnosis and to identify the presence of functioning thyroid tissue in the neck.

A 13-Year Clinical Audit

Eighteen patients with clinical diagnosis of TDCs managed in the Department of Otolaryngology of Universiti Sains Malaysia over 13 years, from 2000 to 2013 were retrospectively reviewed. The clinical data was obtained from the medical record department of the hospital.

Out of the total number of 18 patients, 61.10% were males and 38.90% were females. Most patients presented with anterior neck swelling (83.30%). Only few patients were with either discharging fistula (5.60%) or both anterior neck swelling and abscess (11.10%). The age range at presentation noted was from 9 months to 33 years; with the commonest age of presentation being 11-20 yrs (44.40%) followed by less than 10 years (27.80%) and the least was in the age ranged between 21-30 yrs (11.10%) (Figure 1). Histopathological examination (HPE) was diagnostic of TDC in about 94% and infected branchial cyst (5.60%) as against fine needle aspiration cytology (FNAC) which was detected TDC in about 77%, simple benign cyst (11.10%) and 'others' (11.10%) such as minor salivary gland and chronic inflammation. Clinical imaging using ultrasonography which was consistent with TDC was noted in about 61.10% of the patients whereas 16.70% revealed other diagnosis such as epidermoid cyst and lymph node. Most patients (83.30%) never had any thyroid scan. Furthermore, in more than 22% of the patients, they had no imaging done. Sistrunk operation was the main operative technique carried out

in all our patients. No surgical complications or recurrence was recorded in this series.

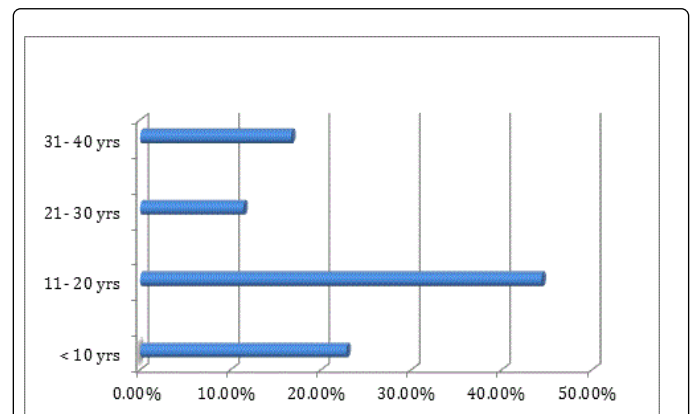


Figure 1: Age distribution of the patients.

Presenting complaint	Percentage
Anterior neck swelling	83.30
Discharging fistula	5.60
Both anterior neck swelling & abscess	11.10
Total	100.00

Table 1: Mode of presentation.

Pathologist's Perspective

Recurrence and the potential for malignancy are two main clinical problems of the TDCs [3]. FNAC can effectively distinguish between benign and malignant. However, in some conditions it cannot give a definite diagnosis due to limited cellularity, reactive changes or cellular degeneration [4]. The common cytomorphologic features of TDC include macrophages, colloid, proteinaceous fluid, cholesterol crystals, lymphocytes, neutrophils, columnar cells, squamous cells (mature or metaplastic type) and rarely thyroid follicular cells [5-7]. Since the TDC was closely related to the thyroid gland development, the cytologic findings were similar to those of thyroid cyst. However, the presence of squamous epithelial cells may be the only significant

cytomorphologic difference between TDC and thyroid cyst as it was not usually noticed in thyroid cyst [5]. But, the squamous cells aspirated were also possibly due to contamination from skin or trachea [8].

Investigation/Results	FNAC	HPE
Thyroglossal duct cyst	77.80	94.40
Simple benign cyst	11.10	-
Infected branchial cyst	-	5.60
Others	11.10	-
Total	100.00	100.00

Table 2: Fine needle aspiration cytology & histopathological examination findings.

USG	Percentage
Thyroglossal duct cyst	61.10
Other diagnosis	16.70
Not done	22.20
Total	100.00

Table 3: Ultrasonography results

Reported incidence of malignancy in TDC is rare and make up less than 1% [9]. A diligent search for the presence of malignant cells is necessary due to hypocellularity that results from dilution by the cystic contents. Carcinoma of the TDC may arise from the cyst lining or from normal thyroid tissue present in the wall of the cyst. Papillary carcinoma was the most common reported comprising 75%- 85%, followed by mixed papillary/follicular carcinoma and squamous cell carcinoma constituted approximately 5% which tended to have the worst prognosis [10]. The diagnostic criteria of papillary carcinoma include high cellularity, follicular cells with intranuclear pseudoinclusion and grooves, powdery chromatin, definite nuclei and psammoma bodies [11]. Squamous cell carcinoma in TDC was suspected when presence of atypical squamous cells admixed with normal follicular cells at the classic midline location. However, most of the cases obtained the definite diagnosis post-operatively. FNAC is still the most reliable method for pre-operative planning to detect a malignant process in midline neck masses and should be considered in all patients except children [12]. A suspicious or definite diagnosis of malignancy by FNAC allows the surgeon to plan the optimum surgical procedure.

Surgeon's Perspective

Operative management

Preoperative assessments for TDC are almost similar to those with other thyroid masses. They include cervical and neck radiograph, ultrasound of the neck or CT scan and thyroid function test. Besides delineating the tract, ultrasound and CT scan are useful to evaluate signs of coincidental malignancies such as mural nodule, calcification or lymph node metastases [13].

Historically, the first operation for TDC was introduced by Schlange in 1893 where the central portion of the hyoid bone was removed in continuity with the cyst. However this technique caused significant recurrence rate approximately 20% [14]. The standard treatment of the operation nowadays follows the technique introduced by Walter Ellis Sistrunk in 1920. He advocated the additional removal of a core of tissue until the submucosal plane at foramen caecum at the base of the tongue [13].

Sistrunk operation has reduced the recurrence rate to 0-5% [13-15]. Risk of recurrence is associated with failure to completely excise the tract following the surgical principles of Sistrunk procedure, presence of multiple tracts around the hyoid bone and wrong diagnosis from initial to other midline cyst such as dermoid cyst.

Recurrent cyst or failed Sistrunk operation

The risk of recurrence is high in case of inadequate tissue resection from the base of the tongue with presence of such multiple tracts. Revision surgery is recommended in view to achieve adequacy of tissue excision. Few approaches are advocated including additional removal of hyoid bone at least two-thirds of the mid-portion, further wide excision of central suprahyoid tissue up to base of tongue mucosa and extended local excision of infrahyoid tissue down to the level of thyroid isthmus [15]. These approaches aim to excise the tract as maximum as possible superiorly without entering oropharynx, remove perihyoid tracts which may present and to excise any possible infrahyoid tract extension to the thyroid gland. If the presence of a malignant lesion can be excluded, percutaneous ethanol injection may be considered a secondary treatment only in selected cases [13].

Incidental carcinoma on thyroglossal cyst

Few factors have been discussed to determine the best treatment modalities and prognosis in incidental carcinoma on TDC. The factors include age of more than 45 years, past radiation exposure, presence of tumour in the thyroid gland on radiological evaluation, presence of clinical or radiological nodes, tumour size of more than 1.5 cm in diameter, cyst-wall invasion, and positive surgical margins on histopathological examination [15]. Histopathological type of tumour like squamous cell or anaplastic will give poorer prognosis. Those who had initial Sistrunk procedure have better prognosis than simple cystectomy [13].

In term of surgical management, it is widely debated whether Sistrunk procedure alone is sufficient to treat. Few studies in which the patient belongs to low risk group showed no recurrence after Sistrunk procedure [15]. Most literatures advocate minimum of total thyroidectomy as treatment of choice in view of possible multifocality of the tumour as well as clear surgical margin from Sistrunk procedure alone [13,15,16]. When nodal metastases are detected preoperatively, total thyroidectomy and regional neck dissection are recommended in addition to Sistrunk procedure. Prophylactic lateral neck dissection in the absence of detectable nodal metastases is not routinely recommended [13]. Radioactive iodine and thyroid suppression therapy are also given in differentiated type of tumour. External beam radiotherapy gives benefits to those belong to squamous cell type [13].

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