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Comparison of thyroglossal duct cysts in adults and children: 11 years of clinical experience

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Cover figure. Axial CT image of the mass in the midline of the neck located anterior of the thyroid cartilage, 4 cm in diameter, compatible with thyroglossal duct cyst. White arrow: thyroglossal duct cyst; Black arrow: macrocalcifications in the cyst.

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Summary

Objective. The aim of this study is to analyse the diagnostic, therapeutic, and follow-up characteristics of patients with thyroglossal duct cysts (TGDC) focusing on clinical differences between paediatric and adult cases.

Methods. Between January 2006 and December 2016, patients who underwent surgical treatment in our clinic with the diagnosis of TGDC were included. Patients were divided into paediatric (under 18 years) and adult groups. Age, gender, presenting symptoms, duration of symptoms, cyst size and location, imaging methods, histopathological findings and recurrence information were compared.

Results. A total of 36 patients were included (17 paediatric, 19 adult). In both groups, the most common complaint was neck swelling, and cysts were most frequently infrahyoid (77.8%). Adults had significantly larger cysts than children ($p = 0.02$), and sinus tract discharge occurred only in paediatric patients ($p = 0.04$). TGDC carcinoma was identified in only 3 adults.

Conclusions. TGDCs are the most common congenital neck masses in both age groups. The gold standard surgical treatment for this condition is the Sistrunk procedure, which significantly reduces the risk of recurrence. Clinicians should remain vigilant, as carcinoma may coexist with TGDCs, particularly in adults.

Keywords: thyroglossal duct cyst, children, adult, comparison, papillary carcinoma

Introduction

Thyroglossal duct cysts (TGDC) are the most common congenital anomalies of the neck which arise from thyroglossal duct epithelium remnants [1]. They appear as painless, soft, partially mobile masses in the midline of the neck and occur in both genders equally [2]. Although they appear more often in children and young adolescents, they can also be diagnosed after 20 years of age because of their asymptomatic clinical course [3]. Due to the inadequate number of comparative studies in the literature, the clinical differences between adult and paediatric patients are not yet clearly defined. In this retrospective study, we aimed to investigate the clinical differences between children and adult age groups by investigating the treatment and follow-up characteristics of patients who were treated with a diagnosis of TGDC between 2006 and 2016 in our clinic.

Materials and methods

Our study was initiated after obtaining approval from the Dokuz Eylul University Non-Invasive Research Ethics Committee (approval number: 2017/09-35). Patients who underwent surgery with a diagnosis of TGDC in Dokuz Eylul University, Department of Otorinolaryngology Head and Neck Surgery between January 2006 and December 2016 and who had at least one-year follow-up were included in the study. Data were collected retrospectively including patient age, gender, complaints and duration of complaints, cyst size and location, radiological methods, surgical methods and complications, histopathological results and recurrence. Imaging studies were obtained at the discretion of the treating clinician. Ultrasound (US) was the first-line modality, with cross-sectional imaging (computed tomography [CT] and magnetic resonance imaging [MRI]) only ordered in selected cases based on clinical judgement rather than a standardized protocol. Statistical analyses were performed with SPSS 22.0.0 (SPSS Inc., Armonk, New York) program. Continuous variables were compared using the Mann-Whitney U test, while categorical variables were analysed using the Chi-square or Fisher's exact test, as appropriate. A p value of less than 0.05 was considered statistically significant.

Results

A total of 36 patients who were diagnosed with TGDC at our clinic between January 2006 and December 2016 were included in the study. Seventeen patients younger than 18 years were included in the paediatric group, and the remaining 19 patients were included in the adult group. Thirteen patients (36%) were females and 23 (64%) were males with a male/female ratio of 1.7. There was no significant difference in terms of gender distribution between the paediatric and adult groups ($p > 0.05$). Age distribution exhibited a bimodal pattern with an overall mean age of 25.1 years; mean age was 6.1 years in children and 42 years in adults. Demographic data and clinical characteristics of the 2 groups are summarised in Table I.

The most common symptom was swelling in the neck which was the main complaint of all patients in both groups. Four of the paediatric patients had discharge on the cutaneous sinus tract (24%), whereas none of the adult patients had discharge ($p = 0.04$).

In both paediatric and adult patients, infrahyoid location was the most common cyst location, with rates of 70.5% and 84%, respectively. There was no statistically significant difference in cyst locations between 2 groups ($p > 0.05$). Cyst locations are summarised in Table I.

The mean cyst diameter among all patients was 19.3 mm ranging from 10 to 40 mm. Cysts were significantly larger in adults than in children, measuring a mean of 22.7 mm (range 10-40) in adults and 15.4 mm (range 10-30) in children ($p = 0.02$).

All patients underwent at least one preoperative imaging study. Ultrasound was the most commonly used modality in both age groups, whereas cross-sectional imaging was performed in a subset of patients, and more frequently among adults. CT was largely confined to the adult cohort, in part to avoid exposing paediatric patients to unnecessary radiation (Fig. 1). Given the retrospective and non-standardised nature of imaging decisions, these data represent observed utilisation patterns rather than an assessment of diagnostic justification. Imaging studies in 2 groups are compared in Table II.

Fine needle aspiration biopsy (FNAB) was performed in 4 of the adult patients preoperatively because of the presence of solid parts in the cysts, and all cytopathological evaluations

Table I. Comparison of demographic and clinical data between children and adults.

	Total (n = 36)	Adults (n = 19)	Children (n = 17)	P
Gender				
Male	64% (n = 23)	63% (n = 12)	65% (n = 11)	p > 0.05
Female	36% (n = 13)	37% (n = 7)	35% (n = 6)	
Age (years)				
Mean	25.1	42	6.1	-
Range	4 mon-67 years	19-67 years	4 mon -14 years	
Complaints				
Swelling	100% (n=36)	100% (n=19)	100% (n=17)	p > 0.05
Sinus/discharge	11% (n=4)	0% (n=0)	24% (n=4)	**p = 0.04**
Duration of complaints (months)				
Mean	19.4	18	21	
Range	1-120	1-120	2-72	p>0.05
Cyst location				
Infrahyoid	77.8% (n = 28)	84% (n = 16)	70.5% (n = 12)	
Suprahyoid	19.4% (n = 7)	16% (n = 3)	23.5% (n = 4)	p > 0.05
Suprasternal	2.8% (n = 1)	0% (n = 0)	6% (n = 1)	
Cyst diameter (mm)				
Mean	19.3	22.7	15.4	**p = 0.02**
Range	10-40	10-40	10-30	
Hospitalisation (days)				
Mean	4.1	5	3.1	p > 0.05
Range	1-17	1-17	1-7	
TGDC carcinoma rate	8.3% (n = 3)	15.8% (n = 3)	0% (n = 0)	p > 0.05

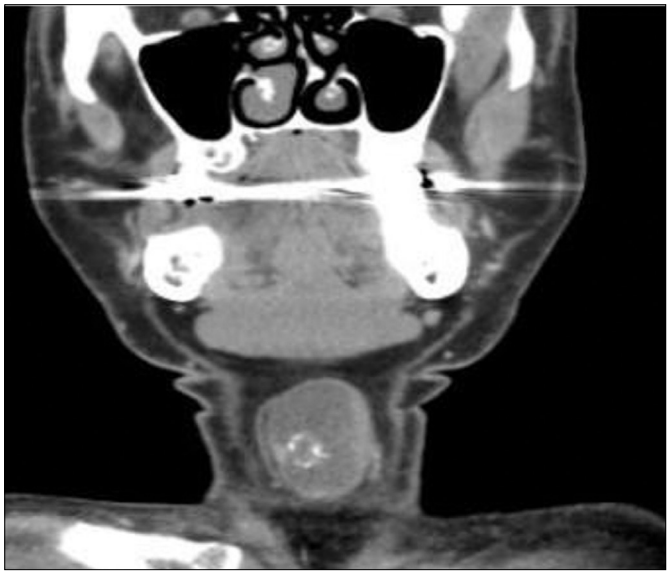


Figure 1. Coronal CT image of the mass in the midline of the neck located anterior of the thyroid cartilage, 4 cm in diameter, compatible with TGDC. The cyst contains opacities like macrocalcification cystic and solid areas, as already seen in the Cover figure.

yielded nondiagnostic results. Preoperative FNAB was not used in any of the paediatric patients. Although FNAB was only used in adults, no statistically significant difference was found for FNAB between groups (p > 0.05).

All patients underwent Sistrunk operation under general anaesthesia which involves excision of the cyst and part of the hyoid bone body by tracking the fistula tract all the way to the foramen caecum. While all the surgical specimens of the paediatric patients were histopathologically consistent with TGDC, 3 of the specimens from adult patients exhibited foci of papillary thyroid carcinomas in addition to TGDC. These 3 patients had undergone FNAB in the preoperative period and none revealed any evidence of malignancy. The cyst diameters of these patients were 30, 30, and 38 mm, respectively, and the diameter of the tumour foci found in the histopathological examinations were 4, 12, and 20 mm respectively. While the incidence of TGDC carcinoma was higher in the adult patient group there was no statistically significant difference between groups (p > 0.05). TGDC carcinoma was detected in 8.3% of all patients and in 15.8% of adult patients (Tab. III).

Table II. Comparison of imaging studies performed in adults and children.

	Adults (n = 19)	Children (n = 17)	Total (n = 36)
US	12 (63.2%)	16 (94.1%)	28 (77.8%)
CT	10 (52.6%)	6 (35.3%)	16 (44.4%)
MRI	3 (15.8%)	0 (0%)	3 (8.3%)
US alone	6 (31.6%)	11 (64.7%)	17 (47.2%)
CT alone	6 (31.6%)	1 (5.9%)	7 (19.4%)
MRI alone	1 (5.3%)	0 (0%)	1 (2.8%)
US+CT	4 (21.1%)	5 (29.4%)	9 (25%)
US+MRI	2 (10.5%)	0 (0%)	2 (5.6%)

The average time of hospitalisation after surgery was 4 days and no statistically significant difference was found between the paediatric and the adult groups ($p > 0.05$). None of the patients experienced any complications in the early postoperative period.

None of the patients had recurrence of TGDC after a mean follow-up of 4.9 years (range 1-12 years).

Discussion

TGDC are the most common congenital neck masses and are seen as painless, semi-mobile, soft swelling in the midline of the neck [4]. Although TGDCs are often asymptomatic, they may manifest as painful, hyperaemic, inflamed masses when infected (Fig. 2). While some studies reported that TGDC is more frequent in the paediatric population, other studies have suggested adults have a higher incidence [5].

Although TGDCs are defined as congenital masses, they are seen, diagnosed and treated in both adults and children. The number of studies comparing the clinical characteristics and surgical outcomes of TGDC between childhood and adulthood is very limited in the literature [3,5].

There were 17 paediatric and 19 adult patients in our series. The male-to-female ratio was 1.7 in the entire group, with a ratio of 1.8 in the paediatric group and 1.7 in the adult one. Several studies in the literature suggest that TGDC is seen equally between men and women, irrespective of age [3,4]. Thompson et al. [6] reported more frequent occurence of TGDC in males during childhood, and a preponderence of females in adults in their large TGDC case series of 685 patients. In our study, with fewer patients, TGDC was seen more frequently in males in both groups and this was slightly more prominent in the childhood.

TGDCs are classified according to their relation to hyoid bone, as intralingual, suprahyoid, infrahyoid, and suprasternal. TGDCs occur predominantly (85%) in suprahyoid and infrahyoid regions, while intralingual and suprasternal presentations are comparatively infrequent (15%) [7]. In our study, cysts were found to be more frequent in the infrahyoid region in both the paediatric and adult groups, which is compatible with the literature [5,8]. The infrahyoid region is followed by the suprahyoid region in both groups and there was no significant difference between the groups in terms of the location of cysts.

In our study, the mean cyst diameter was 19.3 mm and ranged from 10 to 40 mm. The cyst diameter in adults was found to be significantly larger compared to the paediatric group ($p = 0.02$). In a case series of 46 patients Pradeep and Jayashree [3] also reported larger cyst size in adults, but there was no significant difference between groups ($p = 0.06$).

Imaging studies are essential for confirming the diagnosis of TGDC and for assessing lesion extent. US is the primary mo-

Table III. Characteristics of patients diagnosed with TGDC carcinoma (papillary thyroid carcinoma) as a result of the final cytopathological examinations.

	Patient 1	Patient 2	Patient 3
Age/gender	54 / female	54 / male	56 / male
TGDC diameter (mm)	30	30	38
FNAB findings*	Benign cytology	Degenerated cells	Benign cytology
Carcinoma diameter (mm)**	4	12	20
Additional treatment	Total thyroidectomy + RAI	Left hemilobectomy + isthmusectomy	No additional treatment
Duration of follow-up (years)	5	7	2
TGDC recurrence	no	no	no
Papillary thyroid carcinoma recurrence***	no	no	no

** Fine needle aspiration biopsy findings in preoperative period; ** Diameters of papillary thyroid carcinoma foci detected on histopathological examination of TGDC; *** Recurrence of papillary thyroid carcinoma in the neck or thyroid gland; RAI: Radioactive iodine (I-131) therapy.*

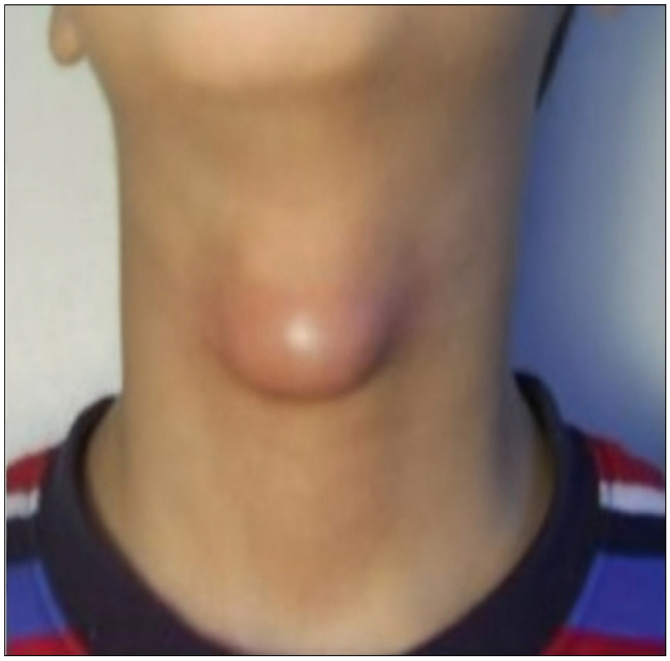


Figure 2. Infected thyroglossal duct cyst in a paediatric patient with swelling in the midline of the neck and hyperaemia of the skin.

dality due to its accessibility, safety, and cost-effectiveness, while CT or MRI is reserved for selected cases requiring detailed anatomical evaluation [9,10]. In our series, all patients underwent at least one imaging study. US was most frequently used in both groups, particularly in children, whereas CT was more often applied in adults. Imaging choices largely reflected clinician preference rather than a standardised protocol.

All of our patients had complaints of swelling in the neck. In the paediatric group, 4 cases of neck cutaneous sinus with discharge were the accompanying complaint. Cutaneous sinus tracts were only observed in paediatric patients and none had an interventional procedure (puncture, incision etc.) performed on the cyst causing the cyst to discharge. The occurrence of cutaneous sinus tracts exclusively in the paediatric group may be attributed to their increased susceptibility to recurrent infection, which frequently results in spontaneous discharge [11].

It may be expected that the duration of the complaints in the adult group with TGDC was longer due to their congenital origin. However, it should not be forgotten that TGDC may be asymptomatic for a long period of time. The mean duration of complaints in our study was 21 months in children and 18 months in adults and there was no significant duration difference between groups, supporting that the TGDC can be asymptomatic for a long time.

Today, the accepted surgical approach for TGDC treatment is the Sistrunk operation. The Sistrunk procedure involves excision of the cystic mass along with the central portion of the hyoid bone body. The fistulous tract is then meticulously followed toward the base of the tongue and removed together with the surrounding soft tissue to reduce the risk of recurrence. A Penrose drain was routinely inserted into the operative site for all patients to minimise the risk of postoperative fluid collections. Simple cyst excision is no longer the preferred surgical method in the treatment of TGDC, as recurrences may occur in up to 50% of patients. The Sistrunk operation reduces recurrence rates to 1-4% [6,7,12]. This procedure was performed in every patients included in the study, and no recurrence was observed after a mean follow-up of 4.9 years.

TGDC carcinoma is a very rare clinical entity which constitutes about 1% of all TGDC cases [13-15]. It is stated that TGDC carcinoma is seen on average around the age of 40. The most common malignancy arising from TGDC is papillary thyroid carcinoma (81.7%) followed by papillary-follicular mixed thyroid carcinoma (6.9%), squamous cell carcinoma (5.2%), follicular carcinoma (1.7%), adenocarcinoma (1.7%), and anaplastic carcinoma (0.9%), respectively [14,16,17].

In our series, papillary thyroid carcinoma was identified in 3 adult patients (15.8%), corresponding to 8.3% of the overall cohort, while no carcinoma was detected in paediatric cases. No significant difference was observed between age-groups ($p > 0.05$). Although this incidence appears higher than reported in the literature, the small sample size limits the statistical power.

FNAB was performed in 4 adult patients with suspicious imaging features; 3 were later confirmed to have carcinoma, yet none were identified preoperatively by FNAB. This reflects the limited sensitivity of FNAB for TGDC carcinoma, consistent with published high false-negative rates of up to 47% [15,17].

Management of TGDC carcinoma remains controversial, with some authors recommending total thyroidectomy [16-19] and others [14,15,20] supporting conservative follow-up in selected patients. In our series, treatment was individualised according to clinical and radiological findings, suggesting that pathological changes in the thyroid gland may represent an important factor in guiding the decision for thyroidectomy.

All of the patients diagnosed with TGDC carcinoma in our study were in the adult group. Although paediatric patients with a diagnosis of TGDC carcinoma have been reported in the literature as young as 6.5 years of age, TGDC carcinoma is extremely rare in children and was not seen in any paediatric patients in our series [21].

Our study has several limitations. The relatively small sample size reduces the statistical power and may limit the generalisability of the findings to broader populations, particularly for rare outcomes such as TGDC carcinoma. The single-centre, retrospective design also carries potential risks of non-standardised medical records, as well as selection bias. In addition, the follow-up period was relatively short in certain cases, which may have limited the detection of recurrences. Further studies including larger case series comparing paediatric and adult patients are needed.

Conclusions

TGDCs are the most common congenital neck masses in both children and adults. Our findings indicate that cysts tend to be larger in adults, whereas sinus tracts are more frequently observed in paediatric patients, underscoring age-related differences in clinical presentation. The Sistrunk operation provided effective treatment in both groups, with no recurrences during follow-up, confirming its reliability across different age populations. Moreover, clinicians should remain vigilant for the possibility of carcinoma, in particular in adult patients.

Conflict of interest statement

The authors declare no conflict of interest.

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Author contributions

TKE, AOI, EO: conceptualisation; OFZ, TKE, ED: methodology; SS, AOI, TKE: project administration; OFZ, ED, EO, MAK: writing-original draft; SS, AOI, TKE: writing-review and editing; SS, EO: data curation; MAK: statistical analysis; SS;AOI, TKE: supervision.

Ethical consideration

This study was approved by the Dokuz Eylul University Non-Interventional Research Ethics Committee (approval number: 2017/09-35).

The research was conducted ethically, with all study procedures being performed in accordance with the requirements of the World Medical Association's Declaration of Helsinki.

Written informed consent was obtained from each participant/patient for study participation and data publication.

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