

Systematic Review Clinical Pathology

Clinical presentation and treatment outcomes of thyroglossal duct cysts: a systematic review

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Abstract. The aim of the present review was to analyze the main clinical signs and symptoms observed in patients with thyroglossal duct cysts (TGDCs). Secondly we investigated the outcomes following the different types of treatment of TGDCs in children and adults. Three selected strings were run on the PubMed database to retrieve articles on these topics. A double cross-check was performed on citations and full-text articles were identified using the study inclusion and exclusion criteria. A meta-analysis was performed of the data obtained. Overall, 356 articles were identified; 24 (comprising a total of 1371 subjects) satisfied the inclusion and exclusion criteria. On the basis of the meta-analysis, the presence of a neck cystic mass was the main clinical presentation of TGDCs, with a mean rate of 75% (95% confidence interval 72–79%). The mean local wound infection rate was 4% (95% confidence interval 3–6%), this being the most frequent complication following treatment. The mean rate of overall recurrence was 11% (95% confidence interval 9–14%). The Sistrunk procedure appears to be the better choice for the therapy of TGDCs to avoid recurrences. Further studies on larger cohorts of patients regarding the minimally invasive treatment options would be helpful to elucidate and endorse their utilization in selected cases.

Key words: thyroglossal duct cysts; Sistrunk procedure; complications; recurrences.

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In head and neck embryology, the thyroid gland descends into the neck as the lateral lingual swellings meet in the midline to form the tongue. During its migration, the middle portion of the gland remains attached to the foramen cecum at the base of the tongue by the thyroglossal duct. This

connection tracks anteriorly to the hyoid bone and usually atrophies around the tenth week of gestation. Incomplete obliteration of the duct gives rise to thyroglossal duct cysts (TGDCs), which are the most common congenital neck mass, with a 7% population prevalence.¹ They gener-

ally present in infants or adolescents, but since they are frequently asymptomatic, some individuals are diagnosed with this defect at age 20 years or older. Both sexes are equally affected.²

The cyst usually presents as a painless, slightly mobile, asymptomatic soft mass,

and most cysts lie in the midline close to the hyoid bone. However, they can be located at any site along the pathway of descent of the thyroid anlage, thus making the diagnosis of TGDC more difficult.³ A variant of TGDC may also be found in the tongue and not present in the neck. These are often discovered incidentally and have been termed lingual TGDCs.⁴

The Sistrunk procedure is the standard treatment for TGDCs in many institutions and consists of the resection of the midline portion of the hyoid bone along with a wide core of tissue belonging to the midline area between the hyoid and foramen cecum. Nevertheless, a number of minor and major complications can be ascribed to surgical mishaps, and the risk of recurrence has not been eliminated.⁵ For these reasons, several authors have proposed variants of the Sistrunk procedure,^{6–8} new medical devices to perform this operation,⁵ and even alternative types of treatment.^{4,9–11}

The objectives of this systematic review of the literature were to analyze the main presenting signs and symptoms of TGDCs and secondarily to focus on the outcomes and possible complications following the different techniques adopted for the treatment of this pathology.

Materials and methods

The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) 2009 guidelines were considered and applied whenever possible in this systematic review. In May 2013, a literature search was performed using the following three search strings in PubMed: (1) (“Thyroglossal Cyst”[MeSH] AND “Signs and Symptoms”[MeSH]); (2) “Thyroglossal Cyst”[MeSH] AND (“complications”[Subheading] OR “Postoperative Complications”[MeSH] OR “Intraoperative Complications”[MeSH]); (3) “Thyroglossal Cyst”[MeSH] AND (“Recurrence”[MeSH] OR “Neoplasm Recurrence, Local”[MeSH]).

The initial search returned a total of 356 results (22 obtained after running the first string, 241 after running the second string, and 93 after running the third string). Abstracts and titles obtained were screened independently by two of the authors (FMG and MR) who subsequently met and discussed disagreements on citation inclusion.

Inclusion criteria for citations were (1) cohort of patients larger than five elements; (2) English language. The exclusion criterion was clear unrelated pathologies.

Of the 356 articles, 29 met the initial inclusion criteria according to both authors (FMG and MR); these were thus obtained and reviewed in detail by the same two authors, who met and discussed disagreements on article inclusion. Inclusion criteria for full text articles and single patients identified were (1) confirmed histological diagnosis of TGDC; (2) cohort of adult population; (3) cohort of juvenile population. Exclusion criteria were (1) reports including cases of TGDC thyroid carcinoma; (2) lack of sufficient information defining exactly the type of treatment used.

A total of seven studies were excluded because there were insufficient data on outcomes (three studies) and treatment techniques (four studies). A further manual check was performed on the references included in the articles and two additional studies were identified that met the inclusion criteria through a review of references and a concurrent PubMed search. The final articles included in the present review were identified ($N=24$), and the main information was extracted and summarized.

By contacting the first authors of five of the studies included in this review by e-mail (Dr Perkins, Dr Hirshoren, Dr Zhang, Dr Simon, Dr Kim), we were able to obtain some missing data which were not reported within these studies.

Statistical methods

Our primary objective was to evaluate the possible clinical presentations of TGDC. The secondary objective was to investigate the different treatment complications and the recurrence rate of the pathology.

We performed a non-comparative meta-analysis, and the heterogeneity between studies was assessed by χ^2 -based Cochran’s Q statistic test and I^2 metric. Heterogeneity was considered statistically significant at $P < 0.01$ for the Q statistic (to assess whether observed variance exceeds expected variance). For the I^2 metric ($I^2 = 100\% \times (Q - df)/Q$), the following cut-off points were used: $I^2 = 0–25\%$, no heterogeneity; $I^2 = 25–50\%$, moderate heterogeneity; $I^2 = 50–75\%$, large heterogeneity; $I^2 = 75–100\%$, extreme heterogeneity. All analyses were performed using Comprehensive Meta-Analysis statistical software, version 2.0 (Biostat, Englewood, NJ, USA).

Results

After an initial check, full-text retrieval, and manual cross-checking of references

included in the articles, 24 studies, comprising a total of 1371 subjects, clearly met the inclusion criteria and were selected for analysis (Fig. 1). The characteristics of these selected studies are included in Table 1.

The majority of the studies were performed with a retrospective cohort design, although three were prospective. The average length of follow-up was reported in only 14 studies (range 1.5–125 months); the overall average length of follow-up was 38.7 months.

Overall, the number of patients in each study included in this analysis varied from 6 to 231. The age of the cohorts varied from a mean age of 3 years to a mean age of 46 years. The vast majority of patients had cervical TGDCs, while an intralingual localization was described in 39 cases.

Concerning treatment modalities (Table 1), of the 1371 patients, 1239 (90.4%) underwent the Sistrunk procedure, either classical or modified, 73 (5.3%) had a simple cystectomy, 19 (1.4%) an endoscopic transoral excision, 17 (1.2%) received ethanol sclerotherapy, 17 (1.2%) OK-432 therapy, and 6 (0.4%) the puncture method.

Clinical presentation and symptoms were described in 16 articles including a total of 1015 patients (Table 2). We analyzed the mean rate of different presentations and symptoms separately: for cervical cystic mass, the mean rate was 0.75 (95% confidence interval (CI) 0.72–0.79) and results were heterogeneous and statistically significant ($I^2 = 86.7\%$; $P = 0.000$); for fistula/draining sinus, the mean rate was 0.18 (95% CI 0.15–0.22) and results were heterogeneous and statistically significant ($I^2 = 78.1\%$; $P = 0.001$); for cervical infection/abscess, the mean rate was 0.34 (95% CI 0.31–0.37) and results were heterogeneous and statistically significant ($I^2 = 92.0\%$; $P = 0.001$); for dysphagia, the mean rate was 0.09 (95% CI 0.07–0.11) and results were moderately homogeneous and statistically significant ($I^2 = 69.8\%$; $P = 0.001$); for airway obstruction, the mean rate was 0.06 (95% CI 0.03–0.09) and results were heterogeneous and statistically significant ($I^2 = 0.0\%$; $P = 0.001$).

The number of treatment complications could be obtained for 22 articles including 1230 patients (Table 3). The rate of total complications occurring in the 22 studies included is illustrated in Fig. 2. Results were homogeneous and statistically significant ($I^2 = 46.5\%$; $P = 0.009$). The mean rate of overall complications was 0.08% (95% CI 0.06–0.10%). We analyzed the single subgroups of complications separately: for seroma, the mean rate was

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