

Management of Thyroglossal Duct Cysts in Children: Retrospective Analysis of a Series of 32 Patients

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Abstract

Original Research Article

Background: Thyroglossal duct cysts (TGDC) are one of the most common congenital anomalies found in the anterior neck region of children. Sistrunk's procedure, described in 1920, is still considered the gold standard. Cyst recurrence is the most feared complication. Our study aims to describe the epidemiological, clinical, and therapeutic features of our patients, as well as reviewing the literature to identify factors that may explain the occurrence of complications and recurrences. **Methods:** The data of 32 patients aged 0 to 16 years who underwent TGDC surgery at Rabat's University Children's Hospital Infant Surgery Department "A", between 2015 and 2020 were retrospectively analyzed. We focused on aspects of clinical presentation, intra- and postoperative treatment, and long-term follow-up. **Results:** This study included 32 cases, out of which five (15,6%) had in their history postoperative recurrences of TGDC (without excising the central part of the hyoid bone), 09 patients had per-operative symptoms of infection, and 4 out of them had intraoperative cyst rupture. Only one out of these four patients had recurrence (3.1%); the only significant risk factor of TGDC recurrence was the non-adherence to the Sistrunk technique. **Conclusion:** TGDC is the most common neck anomaly in children. This condition can be managed successfully without complications and with a low recurrence rate. Preoperative infection and cyst rupture did not influence the recurrence rates.

Keywords: Thyroglossal duct cyst, congenital disease, Sistrunk procedure, complication, recurrence, pediatric surgery.

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INTRODUCTION

Thyroglossal duct cysts (TGDCs) are congenital cervical malformations due to abnormal persistence of the duct resulting from the migration of the thyroid anlage from the base of the tongue to its definitive anatomic location [1]. It is a frequent pathology representing the most common dysembryoplasia in the head and neck [2]. The rate of this disease in the infant population is estimated to be between 70% and 75% of midline swellings of the neck [3, 4]. After adenitis, TGDC is the second most common cause of cervical swelling in children [5]. In the majority of cases, cysts are most commonly discovered in early childhood, but they can appear at any age [6, 7]. Male and female patients are affected with the same frequency [8]. The usual clinical presentation is a cervical mass, movable, non-tender, sometimes elevating during swallowing or protrusion of the tongue [9, 10]. The diagnosis is mainly clinical but ultrasound can be used, which remains the reference paraclinical examination [9]. The evolution is dominated by the risk of occurrence

of episodes of secondary infection responsible for fistulization of these cysts and the risk of malignant degeneration, which is exceptional and only appears in less than 1% of cases [11, 12]. Their management is surgical, the objective of which is to limit the rate of recurrences. Furthermore, the Sistrunk procedure remains the surgical treatment of choice.

METHODS

Study Design:

We conducted a retrospective review of all patients who underwent surgery for TGDC or thyroglossal duct fistula (TGDF) in the Infant Surgery Department "A" at Rabat's University Children's Hospital, between 2015 and 2020.

To safeguard patient privacy, all patient information was de-identified. All patients and their families gave verbal consent to participate, and families gave written permission for publication in cases where

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the results were based on data or photographs. This study's report is compliant with the process guidelines.

All patients under the age of 16 years and who had a confirmed histopathology of TGDC or TGDF were included in this study. Patients with suspicious histopathology for TGDC or TGDF, incomplete results, abnormal thyroid function tests, or neck swelling were excluded for other reasons.

Data Collection

Data collected included patient age, gender, age at TDC presentation, clinical presentation, presence or absence of preoperative infection, imaging modality, surgical management, sizes and locations of cysts, skin fistulization, histologic diagnosis, and histologic presence of inflammation, postoperative infection, complications and recurrence of disease. An exploitation sheet was established as a data collection tool including the various variables necessary for our study which was duly completed using patient records, operating report registers, and Information collected from parents, allowing us to obtain the results that we analyzed as a next step.

Data Analysis

The exploitation of the data statistically made use of the "JAMOVI" software, which enabled us to obtain the results presented in the following chapter.

Intervention

All procedures were performed under general anesthesia in a supine position with an extended neck, after skin preparation and disinfection. The cervical incision was transverse in front of the cyst, well centered in relation to the midline, often concealed in a fold of the neck. In the case of a fistula, this incision circumscribed the fistulous orifice into a wedge of orange. After the dissection of the cyst and the cord up to the body of the hyoid bone to which it adheres, the infrahyoid muscles (mostly sternohyoid and thyrohyoid muscles) were divided. The central part of the hyoid bone was resected along with the included intact tract (Figures 1 and 2). At the end of the surgery, careful hemostasis was ensured, followed by sutures with 2/0 absorbable thread of the sub- and supra-hyoid muscles. The suturing of the superficial planes was done in 2 planes. Postoperative antibiotic therapy was initiated in all patients.



Figure 1: Dissection of the track with adjacent tissues up to the foramen caecum



Figure 2: Cyst of the thyroglossal duct after excision

Data Interpretation

The data were recorded using Microsoft Excel (2019) and a descriptive analysis was carried out using SPSS version 25 (IBM Corp., Armonk, NY, USA).

RESULTS

Among the 32 included in this study, five (15,6%) had in their history postoperative recurrences (simple excising of the cyst) with varying delays from 4 months to 7 years. Only one case (3,1%) of a similar disease in the patient's family was identified. The patient characteristics are summarized in Table 1. 62.5% of the patients were male (n = 20) and 37,5% female (n = 12), giving an M/F ratio of 8/5. The age of the patients at diagnosis and treatment varied from 1 and 15 years with a mean age of 6.6 years. The most common age of presentation was between 6 and 12 years (n = 11). The most common presenting symptom was painless upper midline neck swelling (n =17, 53%), difficulty in swallowing (n = 3, 9.3%), breathing difficulties (n = 2, 6.2%), and in the majority of cases, aesthetic prejudice (n = 19, 59.3%), compel parents to bring their children to a pediatric consultation (Figure 3). The TGDC localization was median in 20 cases (62.5 %), deviated to the left in 7 cases (21.87 %), and to the right in 5 cases (15.6 %). The vertical orientation of the TGDC was specified in 13 patients: supra-hyoid in 8 of our patients (25%), pre-hold in 4 patients (12.5%), and infra-hyoid in 1 patient (3.1%). Physical examination allowed us to identify different clinical presentations of TGDCs: Isolated cervical swelling (n = 21, 65.6%), Cervical swelling associated with infection (n = 9, 28.1%), Fistula associated with inflammation (n = 2, 6.2%), isolated fistula (n = 2, 6.2%). The TGDC's size was mainly larger than 1 cm³ (n = 18, 56.2%), only 5 patients (15.6%) had a cyst smaller than 1 cm³, the size of the remaining group of children (n =9, 28.1%) were not specified. Concerning the rigidity of the cyst at diagnosis, 15 of the patients had

a firm (46.8%), and 5 had a soft (15.6%) neck mass which was not specified in 12 patients. At the time of presentation, 9 patients (28.1%) showed signs of infection (such as warmth, redness, fever, and/or pain) at the TGDC site. There were no signs of infection in the remaining patients for whom this information was given in the database (n = 12, 37.5%). Only one patient had a TGDC recurrence among all patients with preoperative infection (n = 9, 28.1%), but there was no significant difference between the group of patients with a TGDC recurrence and the group of patients without a TGDC who had a preoperative infection.

Those 32 patients were operated on by pediatric surgeons, in a single department 'A' of pediatric surgery. All patients were treated using the Sistrunk procedure. Noteworthy, the five patients with a history of postoperative recurrences, had already their first surgery (simple excising of the cyst, without excising the central part of the hyoid bone); 4 of these 5 patients suffered from an intra-operative cyst rupture (12.5%); and only one patient, who had also a preoperative infection showed a recurrence afterward (3.1%).

All patients received a prophylactic antibiotic treatment during and/or after surgical treatment ranging from 2 to 6 days. The most commonly used products were amoxicillin clavulanic acid.

Abscess development occurred in five cases (15.62%), who did not receive postoperative antibiotics, which progressed well under antibiotic treatment followed by aspiration, with only one case (3.62%) of recurrence being observed. But no significant difference was reached. There were no cases of postoperative hemorrhage, swallowing difficulties, or vocal cord paralysis. The majority of cases were diagnosed with TGDC, followed by TGDF, and there was no sign of malignancy in all cases.

Table 1: Baseline characteristics, operations, and outcomes

Variable	No. of cases (%)
Sex	
Male	20 (62, 5)
Female	12 (37, 5)
Patient age	
1-2	04 (12.5)
2-5	07 (21.8)
5-12	16 (50)
>12	05 (1.5)
Mean age	
Male	6,71 years
Female	6,63 years
Past history:	
Infection	09 (28.1)
Recurrences	05 (15.6)
A similar case in the family	01 (3,1)
Symptoms	
Cervical swelling	17 (53,125)
Aesthetic prejudice	10 (31,125)

Variable	No. of cases (%)
Breathing difficulties	2 (6,25)
Difficulty in swallowing	3 (9,375)
Location	
Median	20 (62.5)
Left paramedian	07(21.8)
Right paramedian	05(15.6)
Supra hyoid	08 (25)
Pre hyoid	04 (12,5)
Infra hyoid	01 (3,1)
Unspecified	19 (59,3)
Physical examination	
Isolated cervical swelling	21 (65.6)
Cervical swelling + infection	07 (21.8)
Fistula + inflammation	02 (6.2)
Isolated fistula	02 (6.2)
Consistency	
Hard	15 (46.8)
Soft	05 (15.6)
Unspecified	13 (40.6)
Ultrasonographic data	
Thyroglossal duct cyst	28 (87.5)
Thyroglossal duct fistula	04(12.5)
Size of TGDCs	
0.1 - 1 cm ³	05 (15.6)
1 - 3 cm ³	08 (25)
> 3 cm ³	10 (31.2)
Unspecified	09 (28.1)
Main operation	
Sistrunk procedure	32 (100)
Outcomes (histopathology)	
TGDC	28 (87.5)
TGDF	04 (12.5)
Postoperative complications	
Infection 05	(15.6%)
Recurrence01	(3.1%)



Figure 3: Typical clinical presentation of a thyroglossal duct cyst

DISCUSSION

TGDC is a congenital neck malformation, with a rate of approximately 7% in the overall population [13], and seems to affect both sexes equally [8]. In our series, we observed a net preponderance of male patients (n = 20, 62.5%). This was also the case in similar retrospective studies [14, 15]. However, other studies

showed an equal affection of both sexes [8, 16, 17] or a majority of female patients [18]. The frequency according to age remains difficult to specify too because some authors have indicated the age of onset of symptoms in their series, while others have reported the age at the time of the initial diagnosis [19]. In our series, the mean age of the female patients at presentation was 6,63 years with extremes ranging from 3 years to 11

years while the mean age of the male patients was 6.71 years with extremes ranging from 1 year to 15 years. This delay in consultation is attributed to ignorance of the disease or negligence, as well as infrastructural and socioeconomic challenges in accessing specialized services. Otherwise, it has been reported in the literature that the duration of symptoms before the patient seeks treatment can range from a few days to 33 years [20]. In addition, it has been found that the cyst of the thyroglossal duct rarely appears before the age of 1-year-old [21]. Surgical removal of the cysts was executed at an average age of 6.6 years. This is contradictory to another study with an average age of 4.52 years [14, 15]. This difference can be explained by the delay in consultation in our context. The cysts found in the neck region of our patients were mostly firm. This is in stark contrast with another study in which the rigidity of the cysts was largely soft [14]. Moreover, the neck mass's dimensions extended one cm³ in the majority of our cases, whereas a previous study showed rather smaller dimensions [22]. TGDC was most commonly found in the midline neck region of the treated patients, which was similar to another retrospective study [8, 15]. It can be seen that if the cyst localization deviated from the os hyoid region, it was mostly towards a suprahyoid or submental position, as shown by a closer look. In contrast, other studies revealed a dominant infrahyoid position [15, 22]. At the time of diagnosis, a preoperative infection was seen in 28.1% of our cases, all of which were treated with antibiotics and/or drainage to cool down the infection. Since a preoperative infection was deemed to be a significant risk factor for TGDC recurrence in previous studies [13, 23], this was also expected in this study, but there were no significantly higher rates of recurrences were noted. On the other hand, some studies reported a correlation between postoperative infection and a higher risk for recurrences than preoperative infections [15, 24]. This could not be confirmed in our study since the five patients with postoperative abscesses did not have a recurrence.

All our patients were treated by surgical excision according to the technique described by Sistrunk comprising dissection of the thyroglossal tract and systematic excision of the body of the hyoid bone and a collar of the muscles of the tongue's base. However, new surgical techniques have been proposed in the literature, such as endoscopy and robotic surgery for the treatment of TGDCs, but, despite their promising results, more multicenter studies are needed to compare head and neck robotic surgeries with other surgical techniques [25-27].

If TGDC recurred, they mostly manifested within the first 4 months after the initial surgery, as also seen in the literature [23]. In addition to previous research, current results showed that simple cyst excision led to a significant increase in recurrences compared to when the Sistrunk procedure was executed. This difference can be explained since the Sistrunk procedure

includes the complete removal of the thyroglossal duct tract by excising the cyst, the central part of the hyoid bone, and its connection to the foramen cecum linguae which in a certain way seems to decrease the chance of redevelopment. Therefore, we can conclude from this study that this almost century-old procedure remains the globally recommended technique in the surgical treatment of TGDC [14, 23, 17, 24].

The use of prophylactic antibiotics showed its role since the development of postoperative abscess was seen in only 5 of the cases (15.6%) who did not take antibiotics postoperatively. 4 of them had an intra-operative cyst rupture (12.5%) of which only one patient (3.1%) had a TGDC recurrence. this cannot be determined as a risk factor for TGDC recurrence. This is contradictory to previous work [28].

Generally, retrospective studies usually include a lot of missing data, as the results section explains, which makes making conclusions more difficult. This data loss was mostly observed for the previously reviewed cases. Therefore, it would be interesting to draft a prospective protocol for data collection to create a broader and more complete view of TGDC recurrence predictors in the future.

CONCLUSION

Although TGDC is the most common congenital neck anomaly in children, it can also manifest in other stages of life. Nevertheless, this condition can be treated effectively with no complications and a low incidence rate of recurrence.

Data Availability Statement: All data and materials are kept by the first and corresponding authors.

Declaration of Conflicting Interests: The authors declare that there is no conflict of interest.

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