

THYROGLOSSAL DUCT CYST, PEDIATRIC PATHOLOGY WITH MULTIDISCIPLINARY MANAGEMENT

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ABSTRACT

Aim of the study The thyroglossal duct cyst is a congenital malformation caused by the persistence of the thyroglossal duct, which connects the root of the tongue with the thyroid. It is the most common cervical cyst, as it occurs in about 7% of the population and constitutes 70% of the congenital malformations that affect the cervical area. Starting with 1920, the gold standard of the treatment of this condition is cyst excision using the described by Sistrunk. Clinical examination is usually enough to set a diagnosis of thyroglossal duct cyst, but confirmation by ultrasound scanning is useful and even necessary to reveal normally or ectopically located thyroid tissue. The primary aim of this study is to describe and present the author's experience with an aparent simple but sometime challenging and multidisciplinary children condition, thyroglossal duct cyst. **Material and Methods** We conducted a retrospective analytical study on 26 children with thyroglossal duct cyst hospitalized in the Pediatric Surgery Department of the “Saint Mary” Emergency Children’s Hospital of Iași for a period of 3 years. **Results** The mean age at the time of examination was 6.4 years, but 53.8% of the children were younger than 6 years. The diagnosis was clinical, confirmed by ultrasound scanning of the soft cervical parts in only 69.2% of the cases. A thyroid ultrasound scanning was performed in only 53.8% of the patients. The thyroglossal duct cyst was surgically removed by the Sistrunk techniques in 84.6% of the patients. Ectopic thyroid tissue was detected in the cyst walls in two of the patients, whereas in another patient the histopathology examination detected cyst invasion inside the hyoid bone (the fifth case reported in literature). The cyst reoccurred in a single patient and it required another surgical procedure. Literature reports a rate of about 1% of malignant thyroglossal duct cysts, usually papillary thyroid carcinoma, and therefore preoperative imaging and histopathology examinations should always be included in the therapeutic protocol. **Conclusions** Clinical examination may be enough to set a diagnosis of thyroglossal duct cyst, but confirmation by ultrasound scanning and laboratory tests is useful and even necessary.

Keywords: thyroglossal duct cyst, child, sistrunk, thyroid.

INTRODUCTION

The thyroglossal duct cyst is a congenital malformation caused by the persistence of the thyroglossal duct, which connects the root of the tongue with the thyroid. During the fetus’ intrauterine life, this duct should normally involve after the seventh week of gestation.

Thyroglossal duct cysts have supra or subhyoid and more rarely suprasternal, intralingual, intrahyoid or mediastinal locations, and since the thyroglossal duct is directly connected with the hyoid bone, the cysts adhere to the hyoid body or horns (1, 2). The persistence of the sole lower section of

the duct materializes in the pyramidal lobe of the thyroid, whereas the upper section of the duct forms the foramen cecum at the root of the tongue. From the histological point of view, the cyst has an inner lining of stratified squamous epithelium or of pseudo-stratified ciliated columnar epithelium. It is the most common cervical cyst, as it occurs in about 7% of the population (3) and constitutes 70% of the congenital malformations that affect the cervical area (4). Its incidence is equal in both sexes. Most patients are children or adolescents whose complaint is a cervical cystic mass, most frequently with medial location, sensitive to palpation; many patients have a recent history of upper airways infection, but it is believed that this has no influence on cyst formation (growth), but increases the probability of cyst detection or may lead to cyst infection. It has the appearance of a round soft well-delimited mass, which adheres to the hyoid and therefore moves with tongue protrusion and deglutition. When it gets infected, the cyst becomes sore and it appears as a cervical suppuration that may open a tegument fistula. When it is big, the cyst may be accompanied by dysphagia, dypnea, dyspepsia due to the pressure applied to the underlying structures (5). The differential diagnosis is set with other medial cervical masses, such as cutaneous abscesses, dermoid cyst, suprahyoid ranula, submental adenitis, lymphoma adenopathy or “cat-scratch disease”, sebaceous cyst, lipoma, branchial cleft cyst.

MATERIAL AND METHODS

We conducted a retrospective analytical study on 26 children hospitalized and treated in the Pediatric Surgery Department of the “Sfânta Maria” Emergency Children’s Hospital of Iași for a period of 3 years (July 2011 – July 2014). The patients were identified according to the disease code entered in the hospital’s computerized database, and then the patients’ records were extracted from the hospital archives. There were 16 girls and 10 boys operated for thyroglossal duct cyst, ages ranging from 1.6 to 14.2 years. The provenience was rural for 18 patients and urban for the rest of them. The patient data

were statistically processed, using the descriptive statistics from the specialized Microsoft Excel data analysis and processing module. We analyzed their demographic data, anamnesis, associated disorders, surgical method chosen, postoperative complications, number of hospitalization days.

RESULTS

In our study 16 patients (61.5%) of the 26 patients with thyroglossal duct cyst were female, and 18 patients of them (69.2%) lived in rural areas. The mean age at the time of examination was 6.4 years (ranging from 1.6 to 14.2 years), but 14 children (53.8%) were younger than 6 years. The reason for their seeing a doctor was the inflammatory phenomena occurring in the anterior cervical area in 12 cases (46.2%). Half of these (23%) initially underwent a simple incision for pus removal and antibiotic therapy and the cyst was surgically removed on their second hospitalization. The remaining 14 children (53.8%) came to the doctor for a pseudotumoral mass in the anterior cervical area, of which only two were sore, whereas the other patients had no other symptoms (fig.1).



Fig. 1. Thyroglossal duct cyst – clinical appearance

The patients’ personal medical history revealed an upper airways infection that preceded the occurrence of the cervical mass in 10 children (38.5%). The diagnosis was clinical, confirmed by ultrasound scanning of the soft anterior cervical parts in only 16 cases (69.2%). A thyroid ultrasound scanning

was performed in only 14 patients (53.8%) which confirmed glandular tissue in normal position. The thyroglossal duct cyst was surgically removed by the Sistrunk techniques in 22 patients (84.6%).

The thyroglossal duct cyst was surgically removed by the Sistrunk techniques in 22 patients (84.6%). The technique was performed by anterior cervical approach and consisted of the removal of the tumor, of the middle section of the hyoid bone and of the cysts extension towards the root of the tongue (fig.2).



Fig. 2. Intraoperative image, view of thyroglossal duct cyst extension towards the root of the tongue

A macro and microscopic histopathology examination of the resection tissue was performed in all the patients (fig.3).

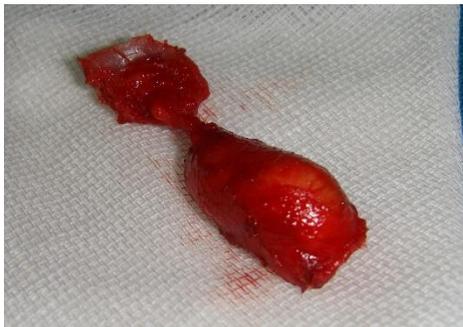


Fig. 3. Resection tissue, view of the hyoid bone section excised with the thyroglossal duct cyst

Ectopic thyroid tissue was detected in the cyst walls in two of the cases (fig.4), but the patients also had normally located glandular tissue (thyroid ultrasonography). In one case, the macroscopic histopathology examination revealed 0.9 cm thick cartilage and bone on a piece of grey 2.5/2.2/1.3 cm tissue, the cross section of which highlighted a 1.2 cm diameter fistula tract (fig.5).

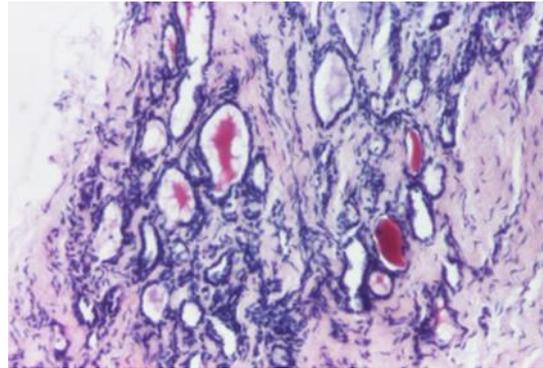


Fig. 4. Thyroid parenchyma in the cyst wall, HE staining, 100x

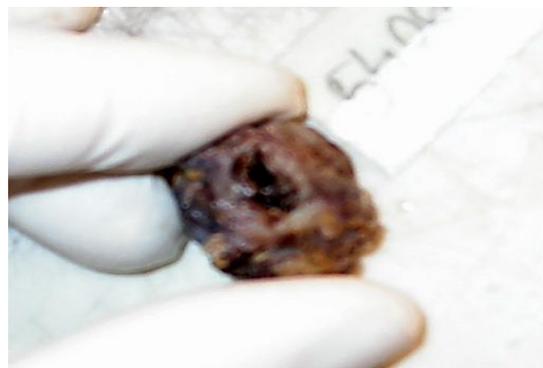


Fig. 5. Thyroglossal duct cyst inside the hyoid one (macroscopic image)

The microscopic examination revealed a cystic structure with lumen lined by newly formed connective-vascular tissue with edema and numerous inflammatory polymorphic elements in addition to hemorrhagic microfoci. The connective wall has rare inflammatory elements. Bundles of striated muscle fibers were detected on the periphery. Pieces of hyaline cartilage and bone made up of bone lamellae and hematogenic bone marrow surrounded by tube-like structures lined by pseudo-stratified epithelium and by stratified squamous epithelium (fig.6, 7, 8); an area with bone lamellae hosting a polymorphonuclear neutrophil (PMN) exudates was also noted.

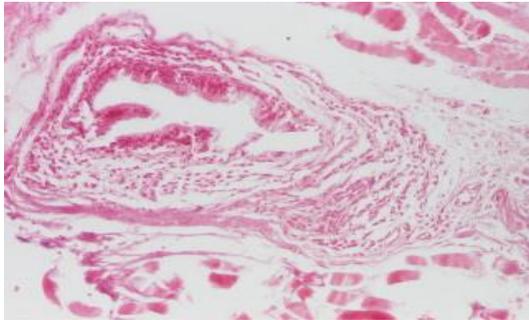


Fig. 6. Tube-like structure lined by a pseudo-stratified epithelium, HE staining, 100x

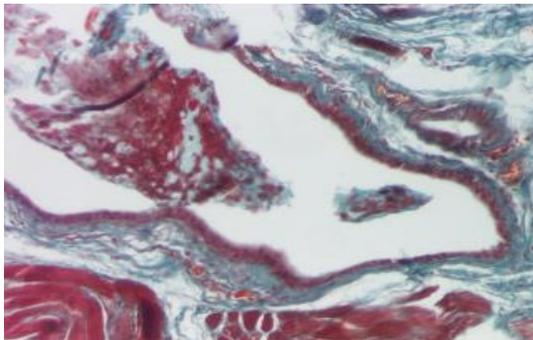


Fig. 7. Tube-like structures lined by a pseudo-stratified epithelium, Szekeley trichromic staining, 100x

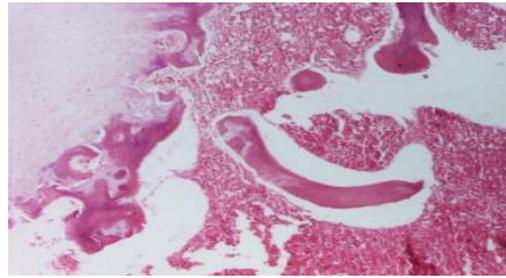


Fig. 8. Hyaline cartilage, bone lamellae and hematogenic bone marrow, HE staining, 40x

The histopathological diagnosis was surgically-removed ulcerated thyroglossal duct cyst with intraosseous duct. An ears, nose and throat (ENT) surgery also attended the surgery in 8 cases (30.7%), and in 16 cases (61.5%) the anesthetist's gesture of pushing the root of the tongue during the resection of the cyst extension towards the foramen cecum was welcome. An incision and cervical abscess draining was carried out in 4 patients (15.4%), and they were rescheduled for the final treatment of their condition at the later date. The cyst reoccurred in a single patient and it required another surgical procedure, although the middle section of the hyoid bone was initially removed. The curative surgical procedure success rate was 95.5%. The average number of hospitalization days for the curative surgical procedure was 4 days (between 1 and 7 days). All the patients returned for follow-up after 10 days, their short-term recovery being positive.

DISCUSSIONS

Thyroglossal duct cyst usually occurs in children under 6 years old in 76% of cases (6), which was also supported by our study, where the mean age on examination was 6.4 years, and 14 children (53.8%) were below 6 years of age. Clinical examination is usually enough to set a diagnosis of thyroglossal duct cyst, but confirmation by ultrasound scanning is useful and even necessary to reveal normally or ectopically located thyroid tissue (7). The ultrasound appearance of a thyroglossal duct cyst is hypoechoic, and any papillary projections, calcifications or a hyperechoic appearance may lead the doctor to suspect a malignant tumor. Thyroglossal

duct cysts may be associated with ectopic thyroid tissue (which is usually inactive) in 25-65% of the cases, despite the thyroid gland being normally located, and in very few cases this thyroid tissue is the only tissue the patient has and its removal leads to hypothyroidism (8, 9). A recent study conducted in England reveals an overall ectopic thyroid tissue incidence of 0.35%. Almost two thirds of these patients (64%) had their ectopic thyroid tissue located inside the anterior cervical mass, which was the only functional thyroid tissue; the incidence of these cases was calculated to be 0.17%. The remaining 36% of patients had their ectopic thyroid tissue located at the root of the tongue (10). Another older study reports an overall ectopic thyroid tissue incidence of 1/100000-1/300000; the ectopic thyroid tissue was located at the root of the tongue in 90% of the cases, or in the anterior cervical area in 10% of the cases (11). Therefore, some therapeutic protocols recommend that, whenever a cervical tumor that requires excision occurs, the patient should first undergo a series of functional tests (TSH, T3, T4), ¹³¹I scintigraphy, or at least thyroid ultrasonography in order to localize the functional thyroid tissue. Thyroid tissue is present in the thyroglossal duct cyst wall in more than 60% of cases, according to Maran's reports (12), but in our study the histopathology examination revealed thyroid tissue inside the cyst only in two patients. Unfortunately, as there were no well established therapeutic protocols in Romania, only 53.8% of our patients underwent thyroid ultrasonography before the operation, which confirmed the normally located glandular tissue, whereas the other patients were exposed to the risk of iatrogenic hypothyroidism.

Literature reports a rate of about 1% of malignant thyroglossal duct cysts, usually papillary thyroid carcinoma (3, 13, 14). None of the patient included in our study group exhibited cellular atypias during the histopathology examination performed, but one should also bear in mind the fact that all our patients were children, unlike other studies that also include adults. There are also studies reporting the occurrence of a

malignant tumor as late as 15 years after the initial removal of the cyst, especially if the excision performed was partial (14). Other possible complications include cyst infection, rupture and recurrence. The latter occurs when the cyst was not completely removed, when there are perioperative infections and especially when the middle section of the hyoid bone or the cyst extension towards the root of the tongue is not removed (15). Cyst infection may cause spontaneous fistula openings in the skin and purulent secretion discharge (16). This is improperly called thyroglossal duct fistula, as there is no communication with the tegument during the fetal intrauterine development.

Although literature reports a success rate as high as 78% for ethanol sclerotherapy of thyroglossal duct cysts (17), starting with 1920, the gold standard of the treatment of this condition is cyst excision using the technique described by Sistrunk. Lesion excision is accompanied by the removal of the middle section of the hyoid bone and of the cyst extension to the root of the tongue, the reported success rate of this procedure being 95-97% (18, 19). A single patient in our study group required another surgical procedure due to cyst reoccurrence, the curative surgery success rate being 95.5% in our clinic. The approach is cervical, by a horizontal incision dissimulated by a natural neck fold. We start by detaching the lower pole of the cyst, then we carefully cut all around it without breaking it, we detach it completely from the adjacent tissues and neck muscles up to the hyoid bone, to which it adheres firmly and the middle section of which should be fastened by two clamps and cut. The dissection continues upwards among the mylohyoid and hyoglossal muscle fibers towards the root of the tongue, in order to isolate the upper extension of the cyst. The removal of this section may be facilitated by the anesthetist's gesture of pushing the root of the tongue by an endobuccal approach. Relapses, consisting of abscesses or fistulae, are possible if the cyst was not fully removed, or if the piece of hyoid it adheres to was not removed. In one patient in the study group the histopathology examination revealed cystic

inclusions inside the hyoid bone, which is extremely rare in literature (2, 20). The intrahyoid location is accounted for by one of the theories of thyroid tissue migration during embryogenesis. An intrahyoid localization is rare and is usually considered as a simple cyst attached to the periosteum. Only four such cases have been reported in literature so far (20). Ghaneim indicated that the cyst was found in between the hyoid bone and the thyroid cartilage in about 60% of the patients and it was suprahyoid, supra-sternal and intralingual in 24%, 13% and 2% of the patients, respectively (1). When the ectopic thyroid tissue is removed with the cervical tumor, either incidentally or expressly, and if the remaining thyroid tissue is insufficient to ensure the euthyroidism state, or if the removed tissue was the only functional tissue, this may be easily identified by a TSH test. The TSH level will exceed 30 μ U/ml within

two weeks after the thyroidectomy. The possible hypothyroidism state will be subsequently followed up by an endocrinologist, who will balance it by levothyroxine administration.

CONCLUSIONS

Clinical examination may be enough to set a diagnosis of thyroglossal duct cyst, but confirmation by ultrasound scanning and laboratory tests is useful and even necessary to reveal normally or ectopically located thyroid tissue. The reported success rate with Sistrunk procedure is 95.5% in our study. The histopathology examination detected one case of intrahyoid cyst invasion, only four such cases being reported in literature so far.

REFERENCES

1. Ghaneim A, Atkins P. The management of thyroglossal duct cysts. *Int J Clin Pract.* 1997; 51(8):512–513.
2. Tas A, Karasalioglu AR, Yagiz R, Doganay L, Guven S. Thyroglossal duct cyst in hyoid bone: unusual location. *J Laryngol Otol.* 2003; 117(8):656–657.
3. Mondin V, Ferlito A, Muzzi E, Silver CE, Fagan JJ, Devaney KO, et al. Thyroglossal duct cyst: personal experience and literature review. *Auris Nasus Larynx.* 2008; 35(1):11–25.
4. Ewing CA, Kornblut A, Greeley C, Manz H. Presentations of thyroglossal duct cysts in adults. *Eur Arch Otorhinolaryngol.* 1999;256(3):136-138.
5. Narayana Moorthy S, Arcot R. Thyroglossal Duct Cyst—More Than Just an Embryological Remnant. *Indian J Surg.* 2011; 73(1): 28–31.
6. Allard RH. The thyroglossal cyst. *Head Neck Surg.* 1982;5(2):134-146.
7. Vulpoi C, Mogoş V, Ciobanu DG, Preda C, Ungureanu MC, Cristea C, Leuştean L, Zbranca E. Thyroid ultrasonography. Interobserver and intraobserver variations. *Rev Med Chir Soc Med Nat Iasi.* 2007;111(4):864-871.
8. Leung AK, Wong AL, Robson WL. Ectopic thyroid gland simulating a thyroglossal duct cyst: a case report. *Can J Surg.* 1995;38(1):87-89.
9. Lilley JS, Lomenick JP. Delayed diagnosis of hypothyroidism following excision of a thyroglossal duct cyst. *J Pediatr.* 2013;162(2):427-8.
10. Joseph J, Lim K, Ramsden J. Investigation prior to thyroglossal duct cyst excision. *Ann R Coll Surg Engl.* 2012; 94(3): 181–184.
11. Tunkel DE, Domenech EE. Radioisotope scanning of the thyroid gland prior to thyroglossal duct cyst excision. *Arch Otolaryngol Head Neck Surg* 1998; 124:597–599.
12. Maran AGD (1997) *Benign disease of the neck.* In: Kerr AG ed, Scott- Brown's Otolaryngology, 6th ed. Oxford: Butterworth- Heinemann, pp 1–19.
13. Dedivitis RA, Guimarães AV. Papillary thyroid carcinoma in thyroglossal duct cyst. *Int Surg.* 2000; 85(3):198–201.
14. Torcivia A, Polliand C, Zioli M, Dufour F, Champault G, Barrat C. Papillary carcinoma of the thyroglossal duct cyst: report of two cases. *Rom J Morphol Embryol.* 2010;51(4):775-777.
15. Perkins JA, Inglis AF, Sie KC, Manning SC. Recurrent thyroglossal duct cysts: a 23-year experience and a new method for management. *Ann Otol Rhinol Laryngol.* 2006;115(11):850-856.
16. Mihoşescu P. Diagnostic errors in persistence of the thyroglossal duct. *Rev Med Chir Soc Med Nat Iasi.* 1967;71(1):203-206.
17. Lee DK, Seo JW, Park HS, Kang MK, Jang AL, Lee JH, Hong JC. Efficacy of Ethanol Ablation for Thyroglossal Duct Cyst. *Ann Otol Rhinol Laryngol.* 2014

18. Cotter ST, Hughes C. Walter Ellis Sistrunk (1880–1933). *Br J Oral Maxillofac Surg.* 2005; 43(2):112-116.
19. Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. *Ann Surg.* 1920; 71:121–122.
20. Bist S, Manisha Bisht, Saurabh Varshney, Nitin Gupta, Rajat Bhatia. Thyroglossal duct cyst in hyoid bone: Unusual location. *Indian J Otolaryngol Head Neck Surg.* 2007; 59(4): 366–368.