Introduction

The involution of the thyroglossal duct typically occurs between 7 and 10 weeks of gestation following migration of the primitive thyroid to its final pre–tracheal position in the inferior neck. Persistence of inferior thyroglossal duct is not uncommon, like presence of the pyramidal lobe of the thyroid gland. Similarly, remnant of upper thyroglossal duct may give rise to cyst formation. Thyroglossal duct cysts are among the most common neck swelling encountered in infancy and childhood. These cysts may develop at any place along the migratory path of the thyroglossal tract of the developing thyroid gland. Most common location is around the hyoid bone superior to the thyroid gland. On review of scientific literature, we found only seventeen documented cases of the TDC within the thyroid gland [1-10]. To the best of our knowledge only one case of intra thyroidal TDC presenting as thyroiditis has been reported [10]. We describe a rare case of intrathyroidal TDC that presented as chronic recurrent pyogenic thyroiditis. According to the PRISMA guidelines, PubMed and Medline were searched for papers from January 1985 to January 2020.In particular, we considered articles with focus on intrathyroidal thyroglossal duct cysts published in English and other languages.

Case report

An otherwise healthy 13- year- old female presented to our head and neck clinic with recurrent episodes of lower neck pain and swelling of 2 years duration. It was associated with repeated episodes of fever and chills & rigors. All the recurrent
episodes were conservatively managed with antibiotics and anti-pyretic medication. There were no signs and symptoms suggestive of hypo or hyperthyroidism. There is no history of thyroid related disorders in the family. Physical examination revealed a 4×3 cm palpable, soft cystic mass in the right thyroid lobe region that moved with swallowing. There were no signs of inflammation or fistula formation on local examination. Laboratory results indicated mild leukocytosis with a white blood cell count of 11,000 cells/μL (normal range is 4,000 to 10,000 cells/μL) and her thyroid function tests were in normal range. Her thyroglobulin antibody, thyroid peroxidase antibody, thyrotropin binding inhibitory immunoglobulin, and thyroid stimulating immunoglobulin levels were normal. Ultrasound demonstrated a 4.3×3.5 cm homogenous low echoic cyst involving mainly the right lobe of the thyroid which extended into the isthmus (Figure 1). Comet tail appearances was seen on ultrasonography due to the calcified debris filling the cyst (Figure 1). Fine needle aspiration yielded thick, viscous, dirty white colored fluid. Cytological examination of the aspirated material revealed lympho-histiocytic tangles, and bland-looking follicular cells. Definitive surgical treatment of right thyroid lobectomy with isthmusectomy was performed. Histopathology reveal a 4.5×3.2 cm cyst involving the right thyroid lobe and isthmus. The cyst was predominantly lined by pseudo-stratified ciliated columnar epithelium with focal squamous metaplasia and mild infiltrate of lymphocytes was seen in the wall (Figure 2 A and B). The TDC was completely surrounded by normal thyroid tissue, with no external tract present. Secondary changes were seen including chronic lympho-histiocytic reaction and dense fibrosis. Histological diagnosis of an intrathyroidal thyroglossal duct cyst was made. Her postoperative period was uneventful. She is being regularly followed up and has not reported any episode of neck pain associated with fever after the surgical procedure.

Discussion

Intrathyroidal thyroglossal duct cyst presenting as chronic thyroiditis is a rare event. Our case represents a unique presentation of the thyroglossal duct cyst in more than one way. First, to the best of our knowledge only 16 cases of the true intrathyroidal TDC’s have been documented in the literature (Table 1) [1–10]. This makes our case 17th case of a true intrathyroidal TDC. Second, only 7 cases of intrathyroidal TDC have been reported in the pediatric age group which makes our case 8th documented case [1,6,9]. Last and rarer, only one case has presented as thyroiditis that too in an adult [10]. This may present either as a goitrous (Hashimoto’s thyroiditis) or a non-goitrous (atrophic thyroiditis) variant.

Thyroglossal duct cysts present mostly in the pediatric and young adults and only one third present after third decade of life. Our review revealed only 7 cases of intrathyroidal TDC in the pediatric age and 9 cases in the adults (Table 1). Most of the cases of the intrathyroidal TDC presented as asymptomatic anterior neck mass, and only in two cases swelling was associated with cases compressive symptoms like discomfort on swallowing (Table 1)[8,10]. Out of the 16 cases only one case exhibited signs of infection and hyperthyroidism (Table 1) [10]. Similar to our case, Barber et al reported a case with history of neck mass and features of thyroiditis [10]. In addition they reported dysphagia, fever, sore throat and hyperthyroidism (Table 1) [10]. On ultrasonography, our case had well defined low echoic cystic nodule involving entire right lobe of the thyroid with calcified debris (Figure 1). Most of the authors have reported hypoechoic cystic nodule in thyroid, while only few have reported complex cysts and one case reported calcified debris, similar to our case (Table 2). It is difficult to distinguish intrathyroidal TDC from other causes of cystic lesions of thyroid gland like colloid cyst, lympho-epithelial cyst and branchial cleft cysts clinically or imaging. While Radionuclide thyroid scintigraphy is useful in cases of lingual thyroid and TDC cysts where a normally located thyroid gland cannot be detected, routine use as part of workup is not justified.

Fine needle aspiration cytology in our case revealed normal looking follicular and squamous cells with lympho-histiocytic tangles. Most of the authors who subjected their cases to the fine needle aspiration reported the presence of normal looking follicular/squamous cells (Table 1). Some authors also reported presence of histio-lymphocytic tangles and macrophages along with normal squamous cells (Table 1). Although, cytology has been shown to have only 62% sensitivity and 69% positive predictive value in the diagnosis of TDCs, our review showed

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Intrathyroidal thyroglossal duct cyst: A rare cause of anterior neck mass, presenting with chronic inflammation and infection

Learning points

- Recurrent episodes of pain and fever associated with swelling in the thyroid region can be signs of intrathyroidal thyroglossal duct cyst.
- Preoperative clinical examination, imaging, FNA are not conclusive of intrathyroidal thyroglossal duct cyst.
- Surgery gives the long term cure by avoiding recurrent infections in the cyst.

Table 1: Intrathyroidal thyroglossal duct cyst: review of literature depicting studies, year of study, age, sex, clinical finding, and FNAC results.

| S.No | Study/Year                | Age/Sex | Clinical Presentation                  | FNAC                              |
|------|---------------------------|---------|---------------------------------------|-----------------------------------|
| 1    | Sonnino, et al. (1989)    | 9/F     | Left thyroid nodule                   | Not mentioned                     |
| 2    | Sonnino, et al. (1989)    | 4/M     | Right thyroid nodule                  | Not mentioned                     |
| 3    | McHenry, et al. (1993)    | 9/M     | Left thyroid mass                     | Normal Squamous epithelial cells  |
|      |                           |         |                                       | Keratinaceous material            |
| 4    | North JF, et al. (1998)   | 78/M    | Thyroid mass midline, Discomfort on swallowing | Normal squamous cells |
| 5    | North JF, et al. (1998)   | 58/M    | Left thyroid mass                     | Normal squamous cells             |
|      |                           |         |                                       | Normal follicular cells           |
| 6    | Hatada, et al. (2000)     | 50/F    | Right Thyroid mass, Discomfort on swallowing | Normal appearing squamous cells   |
|      |                           |         |                                       | No Follicular cells               |
| 7    | Johnston, et al. (2003)   | 10/M    | Left thyroid mass                     | Benign TDC                        |
| 8    | Roy D (2003)              | 50/F    | Right thyroid mass                    | Not Mentioned                     |
| 9    | Perez-Martinez, et al. (2005) | 11/M  | Right thyroid mass                    | Cells suggestive Thyroglossal duct cyst |
| 10   | Choi HJ (2007)            | 41/F    | Left thyroid mass                     | Acellular specimen                |
| 11   | Alvarez Garcia et al. (2015) | 2/M    | Right neck mass                       | Not Mentioned                     |
| 12   | Alvarez Garcia et al. (2015) | 10/M | Right neck mass                       | Squamous epithelium               |
| 13   | Huang LD (2015)           | 45/F    | Bilateral neck mass                   | Not mentioned                     |
| 14   | Saadi R (2015)            | 48/M    | Midline neck mass                     | Benign epithelial cell, macrophages |
| 15   | Barber J (2018)           | 36/M    | Neck mass, Dysphagia, fever, sore throat Hyperthyroidism | Acute inflammation, lympho-histiocytic tangles, Bland follicular cells |
| 16   | Kim, et al. (2018)        | 62/M    | Midline neck mass                     | Benign follicular, squamous cells  |
| 17   | Present case              | 13/F    | Right neck mass, recurrent fever, pain neck | Benign squamous, follicular cells, lymph histolytic tangles |

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### Table 2: Intrathyroidal thyroglossal duct cyst: review of literature depicting study, year of study, ultrasound finding, surgical procedure and histopathology findings.

| Serial No | Study/year               | Ultrasound finding                              | Surgical procedure                                      | Thyroglossal duct tract | Histopathology                        |
|-----------|--------------------------|-------------------------------------------------|---------------------------------------------------------|-------------------------|---------------------------------------|
| 1         | Sonnino, et al. (1989)   | Not mentioned                                   | Cyst excision + Sistrunk procedure                      | Yes                     | Confirmed TDC                        |
| 2         | Sonnino, et al. (1989)   | Not mentioned                                   | Cyst excision + Sistrunk procedure                      | Yes                     | Confirmed TDC                        |
| 3         | McHenry, et al. (1993)   | Not mentioned                                   | Cyst excision + Sistrunk procedure                      | Yes                     | Cyst lined by Keratinizing pseudo stratified squamous epithelium |
| 4         | North JF, et al. (1998)  | Not mentioned                                   | Lobectomy + isthmusectomy                               | No                      | Cyst lined by squamous cells          |
| 5         | North JF, et al. (1998)  | 1.1 cm hypo echoic solid nodule                 | Lobectomy + isthmusectomy                               | No                      | Cyst lined by squamous cells          |
| 6         | Hatada, et al. (2000)    | Low echo intrathyroidal mass                    | Hemithyroidectomy                                       | No                      | Cyst lined by squamous epithelium, surrounded by normal thyroid tissue |
| 7         | Johnston, et al. (2003)  | 3.99 cm Heterogenous-normo-echoic lesions, low echo and fluid parts | Hemithyroidectomy                                       | No                      | Cyst lined by respiratory and squamous epithelium |
| 8         | Roy D (2003)             | 5 cm , complex cyst, left and isthmus           | Hemithyroidectomy                                       | No                      | Cyst lined by respiratory and squamous epithelium, surrounded by normal thyroid tissue |
| 9         | Perez-Martinez, et al.   | 3 cm, cystic with calcified debris              | Hemithyroidectomy                                       | Yes                     | Epithelial lining consistent with TDC |
| 10        | Choi HJ (2007)           | 0.7 cm hypo echoic nodule                       | Hemithyroidectomy                                       | No                      | Cyst lined by non keratinising squamous epithelium |
| 11        | Alvarez Garcia et al.    | 17mm cystic nodule                              | Enucleation                                             | NO                     | Non keratinising. Squamous epithelium |
| 12        | Alvarez Garcia et al.    | 2 cm cystic lesion                              | Enucleation                                             | NO                     | Non keratinising. Squamous epithelium |
| 13        | Huang LD (2015)          | 3.99 cm Normo-echoic focal lesions with low echo and fluid parts | Hemithyroidectomy                                       | No                      | Cyst lined by respiratory and squamous epithelium, surrounded by normal thyroid tissue |
| 14        | Saadi R (2015)           | 1.1 Cystic lesion                               | Isthmusectomy with + Sistrunk procedure                 | Yes                     | Epithelial lining consistent with TDC |
| 15        | Barber J (2018)          | 5 cm , complex cyst                             | Hemithyroidectomy                                       | NO                     | Cyst lined by pseudo stratified ciliated columnar cells, squamous metaplasia, surrounded by thyroid follicles |
| 16        | Kim, et al. (2018)       | 3 cm predominantly cystic mass with calcified debris | Hemithyroidectomy                                       | No                      | Cyst lined by squamous epithelium and pseudo stratified ciliated columnar cells surrounded by thyroid follicles |
| 17        | Present case             | 4.5 cm cystic nodule involving right thyroid lobe and isthmus with calcified debris | Right hemithyroidectomy, isthmusectomy                   | No                      | Cyst lined pseudo stratified ciliated columnar cells with squamous metaplasia surrounded by thyroid follicles. | Lympoh-histiocytic reaction with dense fibrosis. |

### Author contributions

Arsheed Hakeem conceived and drafted the manuscript and prepared the figures. Hassaan Javaid carried out the clinical assessment of the patients. Hima Bindu Rallabandi did the pathology and Fozia wani revised the manuscript for intellectual content.

### Competing interests

The funding organization(s) played no role in the study design; in the collection, analysis, and interpretation of data; in the writing of the report; or in the decision to submit the report for publication.

### Ethical statement

The patient provided written informed consent for publication of the case details and analysis.

### References

1. Sonnino RE, Spigland N, Laberge JM, Desjardins J, Guttmann FM (1989) Unusual patterns of congenital neck masses in children. J Pediatr Surg 24: 966-969. [Link:](https://bit.ly/31nKzJk)

2. McHenry CR, Danisch R, Murphy T, Marty JJ (1993) Atypical thyroglossal duct cyst: a rare cause for a solitary cold thyroid nodule in childhood. Am Surg 59: 223-238. [Link:](https://bit.ly/3g4Xlk8)

3. North LJH, Foley AM, Hamill RL (1998) Intrathyroidal cysts of thyroglossal duct origin. Am Surg 64: 866-888. [Link:](https://bit.ly/2A8E7e5)

4. Hatada T, Ichii S, Sagayama K, Ishii H, Sugihara A, et al. (2000) Intrathyroidal thyroglossal duct cyst simulating a thyroid nodule. Tumori 86: 250-252. [Link:](https://bit.ly/3dGFCDa)

5. Johnston R, Wei JL (2003) Intra-thyroid thyroglossal duct cyst as a differential diagnosis of thyroid. Int J Pediatr Otorhinolaryngol 67: 1027-1030. [Link:](https://bit.ly/2Z7hyyE)

6. Roy D, Roy PG, Malik VK, Seenu V (2003) Intrathyroidal thyroglossal duct cyst presenting as thyroid nodule. Int J Clin Pract 57: 637-638. [Link:](https://bit.ly/3i3A2ca)
7. Perez-Martinez A, Bento-Bravo L, Martinez-Bermejo MA, Conde-Cortes J, de Miguel-Medina C (2005) An intra-thyroid thyroglossal duct cyst. Eur J Pediatr Surg 15: 428-430. Link: https://bit.ly/388sMHr

8. Choi HJ, Jung JH, Yoo J. (2007) Intra-thyroid thyroglossal duct cyst: A case report. Korean J Pathol 41: 132-134.

9. Alvarez Garcia N, Burgues Prades P, Gonzalez Martinez- Pardo N, Simon

Portero S, Fernandez Atuan R (2015) The intra-thyroid thyroglossal cyst in the differential diagnosis of the solitary thyroid nodule: A presentation of 2 cases. An Pediatr (Barc) 82: 360-361. Link: https://bit.ly/2CNdxr

10. Barber J, Martinez DS, Diaz FP, Stark AP, Masha J (2018) Intrathyroidal thyroglossal duct cyst: a rare cause of thyroiditis in an adult. AACE Clinical Case Reports 4: e90-e93.