

Original Article

Intra-thyroid thyroglossal duct cyst: a case report and review of literature

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Abstract: Thyroglossal duct cyst is the most common congenital cyst in the head and neck, which is defined usually occurring in children. However, intra-thyroid thyroglossal duct cyst in an adult is unusually found. Here we describe a case of a 45-year-old woman who was found neck mass along the midline for 5 years. During the surgery we found a separated nodule in the left inferior pole of the thyroid. Surprisingly the diagnosis of the nodule was confirmed by pathology and histological examination demonstrating that it was the thyroglossal duct cyst. Intra-thyroid thyroglossal duct cyst in an adult is a rare finding, with few cases reported. For it is generally thought that any thyroid tissue found in the lateral aspect of the neck may indicate metastatic deposits from well-differentiated thyroid carcinoma. Although pathogenesis of an alone thyroglossal duct cyst in the left inferior pole of the thyroid remains unknown, our case could suggest thyroglossal duct cyst should not be excluded in the differential diagnosis of lateral neck masses especially when it simulates nodules in the thyroid.

Keywords: Intra-thyroid thyroglossal duct cyst (ITTDC), thyroid gland, nodule, ultrasound (US)

Introduction

Thyroglossal duct cysts are the most common congenital mass to arise in the neck [1]. Remnants of the duct account for 70% of all congenital neck anomalies [2, 3]. In most situations, a careful clinical history and physical examination are efficient to make a correct pre-operative diagnosis [4]. However, it sometimes remains difficult whether these nodules represent benign embryological remnants, or whether they represent metastatic disease from primary thyroid carcinoma or thyroid goiter. Here, we report a case of thyroglossal duct cyst in the left inferior pole of the thyroid in a patient that simulated thyroid goiter. In addition, we study previous related researches about thyroglossal duct cysts and make a literature of review.

Case report

A 45-year-old woman presented with a five-year history of bilateral neck mass along the midline. The patient had no significant past medi-

cal, surgical history and was completely asymptomatic. She denied any family history of thyroid disease or history of head and neck irradiation. Physical exam revealed a nontender, mobile and smooth neck mass measuring 4 cm × 4 cm just in the left inferior pole of the thyroid. The right lobe was not palpable. There was no associated cervical lymphadenopathy. The rest of the physical exam was unremarkable. The chest radiography showed no abnormal findings. Blood calcium level was also checked for the possibility of medullary thyroid cancer, and was within normal limits. The patient's thyroid function was normal. Laboratory examinations including thyroid hormone indicated normal. Moreover the thyroglobulin antibody and thyroid peroxidase antibody is < 0.90 IU/ml and 1.50 IU/ml respectively, both of which are in the normal range.

Ultrasonography of the neck revealed heterogeneous thyroid gland with presence of numerous normoechoic focal lesions with low echo and

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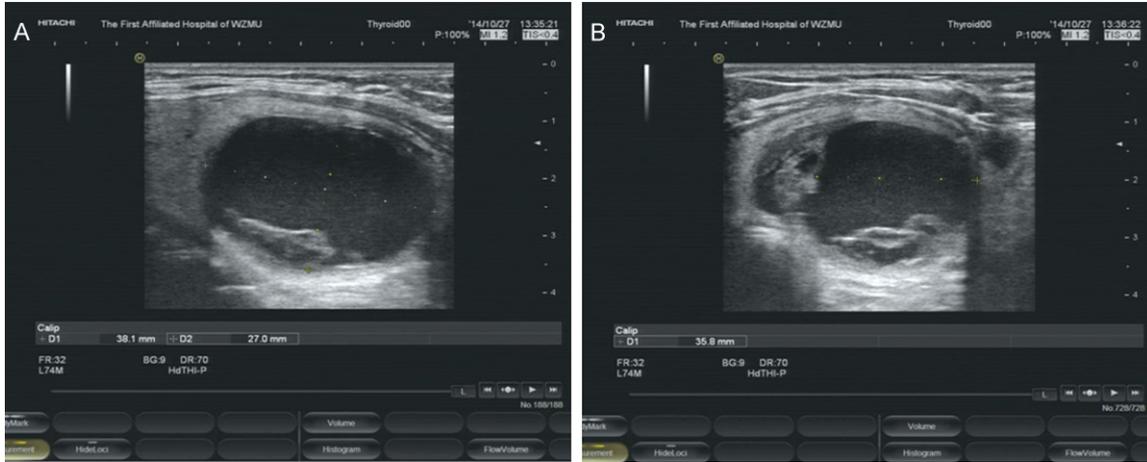


Figure 1. Ultrasonography of the nodule showed heterogeneous thyroid gland with presence of numerous non-mochoic focal lesions with low echo and fluid parts in left lobe with the nodule 3.99 cm × 2.69 cm.

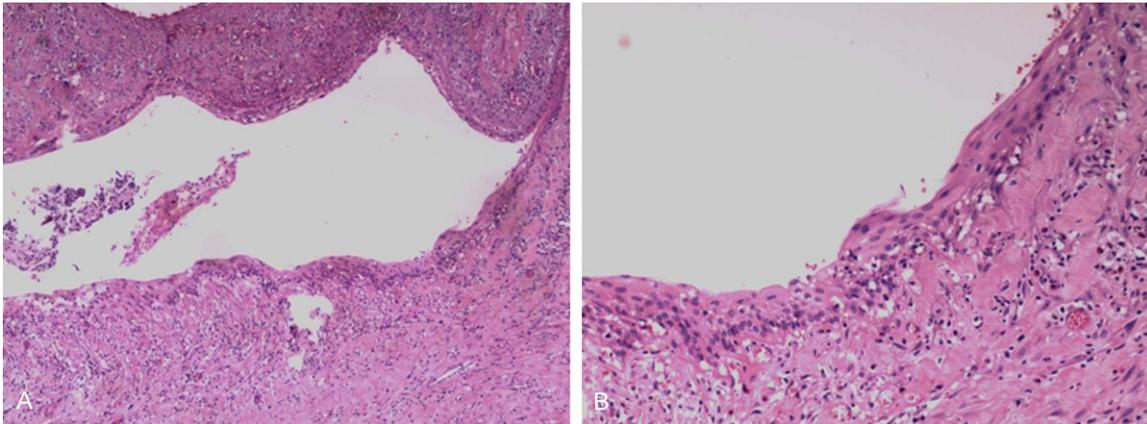


Figure 2. Hematoxylin and eosin (H&E) histology of thyroglossal duct cyst (20 ×) (A) and (100 ×) (B). The pathology examination of the nodule which showed pseudostratified ciliated columnar and squamous epithelial lining associated with thyroid follicles in the surrounding stroma.

fluid parts in left lobe with the nodule 3.99 cm × 2.69 cm (**Figure 1A, 1B**).

The surgery was performed during which we found that a tough and independent cystic nodule measuring 4 cm × 4 cm taking up most of the left inferior pole of the thyroid. The nodule was covered by an intact membrane containing hemorrhagic liquid without calcification while there were no swelling lymph glands detected in surrounding tissues. The frozen sections of nodule were performed before the next treatments. The specimens of the nodule were reviewed in our pathology department, however, surprisingly demonstrating to be thyroglossal duct cyst. And no thyroglossal duct tract was noted. Our patient underwent a left hemithyroidectomy. The patient had no compli-

cations during the postoperative period and the tracheostomic tube was removed on the second day after surgery. Patient went back home.

The diagnosis of thyroglossal duct cyst was confirmed by pathology examination, which showed pseudostratified ciliated columnar and squamous epithelial lining associated with thyroid follicles in the surrounding stroma (**Figure 2**). Referring to the previous reports the criteria for the pathology of thyroglossal duct cysts were as follows: TDCs are lined by stratified squamous or pseudostratified ciliated columnar epithelium with mucous glands and thyroid follicles. As a result they contain mucoid and proteinaceous material. Cholesterol crystals have also been described in TDC [5, 6]. Thus our pathology examination just met with these

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Table 1. Summary of cases of intrathyroidal TDC

References	Age/ Sex	Location	Presentation	Gross Pathology	Other pathologi- cal condition
Choi HJ et al. (11)	41/F	in the left thyroid lobe	asymptomatic swelling	A palpable, firm, asymptomatic and defined 0.7 x 0.6 cm mass	Not mentioned
Hatada T et al. (12)	50/F	in the right thyroid lobe	Visible asymptomatic swelling	A mobile, nontender and clearly defined 1 x 2 cm mass	Not mentioned
Roy D et al. (13)	50/F	in the right thyroid lobe	painless and asymptomatic swelling	a mobile, nontender, well defined 1 x 1 cm mass	Not mentioned

criteria. Histologically, the nuclear features of the thyroid follicles were bland. The nuclei were small, round with no ground-glass nuclei, pseudoinclusions, nuclear grooves, mitosis, papillae, stromal reaction, psammoma bodies, necrosis, or vascular invasion and did not show any morphologic features of papillary cancer. The entire thyroglossal duct cyst was benign. Accordingly, a diagnosis of thyroglossal duct cyst arising in the left inferior pole of the thyroid was established.

Discussion

Thyroglossal duct cysts (TDCs) are the most common congenital neck mass which often occur in pediatric patients, however, at least half are found in the second decade of life and they can also present later in adulthood [7]. It is important to understand the embryological origin and development of the thyroid gland, which would make us acquainted with the etiology of thyroglossal duct cyst. The primordium of the thyroid gland is combined with the tongue by a narrow tubular structure called the thyroglossal duct, the site of whose connection with the epithelial floor of the mouth is signified by the foramen caecum [8]. From here it extends caudally in the midline and ventral to the primordium of the hyoid bone, posterior to which it may structure a recurrent loop. As a consequence the thyroglossal duct cysts usually atrophy and disappear during the eighth and tenth weeks of gestation [9]. Nevertheless if the duct persists, regarding repeated local infection or inflammation, secretion from the epithelial lining may accumulate, triggering cyst formation [10]. However our case of TDC arising from the left inferior pole of the thyroid is intensely rare. A few cases of intrathyroidal TDC were reported in the literatures (Table 1) [11-13]. To our knowledge, this maybe the extremely rare case report of intrathyroidal TDC in adults in China.

Both TDCs and branchial cleft cyst (BCC) can present as thyroid masses [14-19]. The BCCs,

arising from failure of obliteration of the cervical sinus, are usually located in a second branchial groove cyst is high in the lateral neck [20]. The discriminating characteristics between a TDC and BCC involve performance of the mass, with TDC more likely to be midline neck masses and infrequent of infection while BCC are typically lateral neck masses. The most valuable method to differentiate the two is the histology examination, for TDCs have walls composed of alternating non-keratinizing stratified squamous epithelium and respiratory epithelium, while BCC walls are composed of predominantly non-keratinizing stratified squamous epithelium with distinctive subepithelial lymphoid tissue throughout [15, 16, 21]. Also we should bear in mind cytology of the aspirated material may be helpful to distinguish TDCs from a dermoid cyst, lipoma, aberrant thyroid tissue, thymic cyst, plunging ranula and teratomas [20].

Moreover it has reported that malignancy is rarely encountered in TDCs. When such rare tumors do develop which may only account 1% in patients with TDCs, they usually present either papillary carcinoma of thyroid origin, or squamous carcinoma [8]. Criteria for the diagnosis of primary papillary carcinoma arising in a TDC after Widstrom are: 1) histological identification of TDC demonstrating that the cyst or duct has an epithelial lining with normal thyroid follicles in the cyst wall; 2) there is normal thyroid tissue adjacent to the tumor; and 3) histopathologic examination of the thyroid gland reveals no signs of primary carcinoma [22]. However, the histology result of our case was quite unequivocal and we could exclude the possibility of primary carcinoma.

The treatment of choice for TDC is the Sistrunk's procedure. Schlange described a procedure that contained the removal of the midportion of the hyoid bone in continuity with the TDC in 1893 for the first time while in 1920 Sistrunk added to Schlange's procedure the excision of a block of tissue between the hyoid bone and

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the foramen cecum to obtain complete removal of TTRs [23, 24]. Regarding our case, we did not find thyroglossal duct tract so a left hemithyroidectomy was performed. Some cases were reported before that the hemithyroidectomy was performed to the patient with only a separated thyroglossal duct cyst as well as no thyroglossal duct tract [20] and the patients who underwent excision alone did not demonstrate recurrence up to 3 years postoperatively on follow-up [16]. Consequently in our case the cyst was completely embedded within the thyroid gland and no tract was identified, therefore lobectomy was performed and believed to be sufficient.

To sum up, in the case presented here, we describe a patient who appeared asymptomatic with thyroid nodule and turned out to be intra-thyroid thyroglossal duct cyst (ITTDC). TDCs are the most common cause of midline-congenital cyst formation in the neck that may present at any age. Generally, it presents as an anterior midline neck swelling which is mobile with deglutition and protrusion of the tongue. However, non-classic presentation is not uncommon and variability in the site, clinical presentation and radiological appearance must be anticipated [25], which is just like our case. As the possibility of metastatic well-differentiated thyroid carcinoma was strongly taken into consideration and a diagnostic hemithyroidectomy was performed. Confirming by histology examination, it in fact was ITTDC. Thus we suggest the ITTDC should be included in the differential diagnosis of lateral neck masses since it is a very rare situation for thyroglossal duct cyst other than metastatic well-differentiated thyroid carcinoma simulating the thyroid nodule. Recurrent disease in the patient will be monitored owing to the possibility that this is a metastasis remaining. This is a rare case report of intra-thyroid thyroglossal duct cyst with no symptoms and classic presentation.

Disclosure of conflict of interest

None.

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References

- [1] Ward GE, Hendrick JW and Chambers RG. Thyroglossal tract abnormalities, cysts and fistulas; report of 105 cases from the Johns Hopkins Hospital observed during the years 1926 to 1946. *Surg Gynecol Obstet* 1949; 89: 727-734, illust.
- [2] Wu SY and Juler G. Thyroid Disease: Endocrinology, Surgery, Nuclear Medicine, and Radiotherapy. *Thyroid* 1991; 1: 369-369.
- [3] Ellis PD and van Nostrand AW. The applied anatomy of thyroglossal tract remnants. *Laryngoscope* 1977; 87: 765-770.
- [4] Davenport M. ABC of general surgery in children. Lumps and swellings of the head and neck. *BMJ* 1996; 312: 368-371.
- [5] Chow LT, Lee JC, Chow WH and Chan SK. Cholesterol-rich thyroglossal cyst: report of a case diagnosed by fine needle aspiration. *Acta Cytol* 1996; 40: 377-379.
- [6] Noyek AM and Friedberg J. Thyroglossal duct and ectopic thyroid disorders. *Otolaryngol Clin North Am* 1981; 14: 187-201.
- [7] Turkyilmaz Z, Sonmez K, Karabulut R, Demirgoullari B, Sezer C, Basaklar AC and Kale N. Management of thyroglossal duct cysts in children. *Pediatr Int* 2004; 46: 77-80.
- [8] Mondin V, Ferlito A, Muzzi E, Silver CE, Fagan JJ, Devaney KO and Rinaldo A. Thyroglossal duct cyst: personal experience and literature review. *Auris Nasus Larynx* 2008; 35: 11-25.
- [9] Remy HB. The thyroglossal cyst. *Head Neck Surg* 1982; 134-146.
- [10] Johnson IJ, Smith I, Akintunde MO, Robson AK and Stafford FW. Assessment of pre-operative investigations of thyroglossal cysts. *J R Coll Surg Edinb* 1996; 41: 48-49.
- [11] Choi HJ, Jung JH, Yoo J, Kang SJ, Lee KY, Chun CS, Kang BJ and Cha ES. Intra-thyroid Thyroglossal Duct Cyst: A Case Report. *Korean Journal of Pathology* 2007; 41: 132-134.
- [12] Acierno SP and Waldhausen JH. Congenital cervical cysts, sinuses and fistulae. *Otolaryngol Clin North Am* 2007; 40: 161-176, vii-viii.
- [13] Shifrin A and Vernick J. A thyroglossal duct cyst presenting as a thyroid nodule in the lateral neck. *Thyroid* 2008; 18: 263-265.
- [14] McHenry CR, Danish R, Murphy T and Marty JJ. Atypical thyroglossal duct cyst: a rare cause for a solitary cold thyroid nodule in childhood. *Am Surg* 1993; 59: 223-228.
- [15] Josephson GD, Spencer WR and Josephson JS. Thyroglossal duct cyst: the New York Eye and

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- Ear Infirmary experience and a literature review. *Ear Nose Throat J* 1998; 77: 642-644, 646-647, 651.
- [16] Sonnino RE, Spigland N, Laberge JM, Desjardins J and Guttman FM. Unusual patterns of congenital neck masses in children. *J Pediatr Surg* 1989; 24: 966-969.
- [17] Hatada T, Ichii S, Sagayama K, Ishii H, Sugihara A, Terada N and Yamamura T. Intrathyroid thyroglossal duct cyst simulating a thyroid nodule. *Tumori* 2000; 86: 250-252.
- [18] North JH Jr, Foley AM and Hamill RL. Intrathyroid cysts of thyroglossal duct origin. *Am Surg* 1998; 64: 886-888.
- [19] Yoskovitch A, Laberge JM, Rodd C, Sinsky A and Gaskin D. Cystic thyroid lesions in children. *J Pediatr Surg* 1998; 33: 866-870.
- [20] Johnston R, Wei JL and Maddalozzo J. Intra-thyroid thyroglossal duct cyst as a differential diagnosis of thyroid nodule. *Int J Pediatr Otorhinolaryngol* 2003; 67: 1027-1030.
- [21] Karmody CS. Developmental anomalies of the neck. *Pediatr Otolaryngol* 1990; 1497-1511.
- [22] Widström A, Magnusson P, Hallberg O, Hellqvist H and Riiber H. Adenocarcinoma originating in the thyroglossal duct. *Ann Otol Rhinol Laryngol* 1976; 85: 286-290.
- [23] Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. *Ann Surg* 1920; 71: 121.
- [24] Schlange H. Ueber die fistula colli congenita. *Arch Klin Chir* 1893; 46: 390-392.
- [25] Thabet H, Gaafar A and Nour Y. Thyroglossal duct cyst: Variable presentations. *Egyptian Journal of Ear, Nose, Throat and Allied Sciences* 2011; 12: 13-20.