

A Case of Teratoma of Thyroid Gland in Adolescence

Yu-Chang Lee, Su-Jin Jeong, Sol-Jae Lee, Chong-Hwa Kim, Yong-Hoon Lee, Jung-Eun Lee and Hye-Ji Seo

Division of Endocrinology and Metabolism, Department of Internal Medicine, Sejong General Hospital, Bucheon, Korea

Benign teratomas of the thyroid are very rare in adolescence and adults. In this review, we report a case of 14-year-old Korean girl with huge neck mass. She presented with anterior neck enlargement and compression symptom which was rapidly aggravated in 2 months. In physical examination, enlarged and firm right thyroid lobe was palpated and laboratory test of thyroid function was normal. In ultrasonography, right lobe was mainly occupied with a solid nodule size of 44×23×25 mm, showing markedly inhomogeneous hypoechogenicity. As fine needle aspiration cytology was non-diagnostic, lobectomy was done. Histological examination demonstrated that the tumor is benign thyroid teratoma composed of tissue from all three germ layers. When large thyroid nodule is detected in adolescence and malignancy could not be ruled out, final diagnosis should be made with surgical resection. And we should at least attentive for possibility of teratomas when ultrasonographic findings are suspicious.

Key Words: Adolescent, Thyroid, Teratoma, Benign, Lobectomy

Introduction

Teratomas are most common type of germ cell tumor, derived from all three germ cell layer, i.e., mesoderm, endoderm, and ectoderm. Teratomas are dominantly observed in ovary, testis, and occasionally found in sacrococcygeal area, anterior mediastinum, retroperitoneum, head and neck lesion.^{1,2)} Head and neck teratomas predominantly arise in infants and babies and occupy 3% to 6% of all teratomas, with extremely rare cases reported associated with the thyroid gland.³⁻⁶⁾ In these days, various types of thyroid nodules could be found by sensitive techniques such as ultrasonography (US).³⁾ However, thyroid teratoma is an exceptionally rare case, which practitioner might neglect.

In our case, teratoma was hard diagnosis to be attained by ordinary tests like US and fine needle aspiration. We diagnosed a benign teratoma of thyroid by lobectomy in an adolescent patient with nodule larger than 4 cm. Therefore, we would like to report the diagnostic image findings and the necessity for surgery of benign thyroid teratoma through this case.

Case Report

A 14-year-old girl presented with enlarged neck mass and compression symptom and discomfort for 2 months. There were no specific findings in previous medical history especially associated with thyroid. There wasn't remarkable disease history in family.

Physical examination revealed enlarged, firm and non-tender palpable mass on the right side of the

Received February 20, 2017 / Revised 1st March 17, 2017, 2nd March 23, 2017 / Accepted March 23, 2017
Correspondence: Su-Jin Jeong, MD, PhD, Division of Endocrinology and Metabolism, Department of Internal Medicine, Sejong General Hospital, 489 Hohyeon-ro, Sosa-gu, Bucheon 14754, Korea
Tel: 82-32-340-1115, Fax: 82-32-340-1834, E-mail: tusyle@naver.com

Copyright © 2017, the Korean Thyroid Association. All rights reserved.
© This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

neck. She had blood pressure of 102/62 mmHg with heart rate 76 beats per minute, respiration rate 20 breaths per minute, and body temperature of 36.2°C was checked. In laboratory investigation, complete blood count is normal; white blood cell count of 6,060/mm³, hemoglobin of 12.4 g/dL, hematocrit of 36.6%, and platelet count of 327,000/mm³. Other laboratory tests were within normal limit; blood urea nitrogen/creatinine of 11/0.68 mg/dL, aspartate aminotransferase/alanine aminotransferase, alkaline phosphatase of 15/10 IU/L, 54 IU/L, sodium/potassium level of 143/3.8 mmol/L and calcium/phosphate level of 9.0/4.2 mg/dL, Free thyroxine was 1.4 ng/dL (reference range, 0.78–1.84 ng/dL), thyroid-stimulating hormone was 2.1 IU/mL (0.2–5 IU/mL). Antithyroglobulin and antithyroid peroxidase antibody tests were both negative and TSH receptor stimulating immunoglobulin was not detected and calcitonin was 6.6 pg/mL which is within normal range (0–10 pg/mL).

Thyroid US revealed a hypoechoic and heterogeneous nodule measuring 4.4 cm in diameter within intrathyroidal area of the right thyroid lobe. Hyperechoic lesions in this nodule were also observed (Fig. 1). US-guided fine needle aspiration cytology (FNAC) was performed and cytological examination showed cystic liquid without thyroid follicular cells and was non-diagnostic.

Although results of FNAC and US of the neck suggested low possibility for malignant tumor, we could

not rule out malignancy because of her age, the size and rapid growth of mass in short period. Furthermore, she complained progressive compression symptom which could be the sign of malignancy. As the result of frozen biopsy was benign, right lobectomy was performed and oval shaped cystic mass in right thyroid gland is observed with size of 3.8×2.8×2.5 cm in dimensions (Fig. 2). The cut surface demonstrated a well capsulated cystic mass with yellow gelatinous and grumous material admixed with many black colored hairs. Portions of thyroid gland are largely replaced by well encapsulated cystic mass, lined by well differentiated stratified squamous cell epithelium and many skin appendages including hair follicles, hairs, many sebaceous glands and sweat glands. The cystic wall partly contains cartilage and bony tissue, mature fat tissue, smooth muscle, and multiple mucous cystic

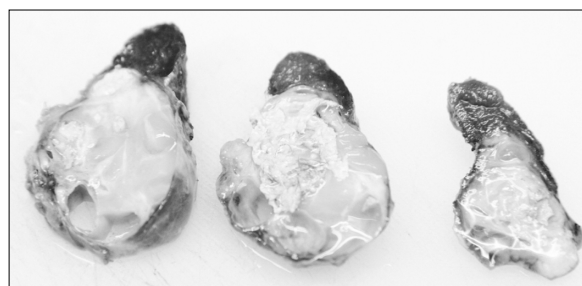


Fig. 2. Cut surface of teratoma demonstrated a well capsulated cystic mass with yellow gelatinous and grumous material admixed with many black colored hairs.

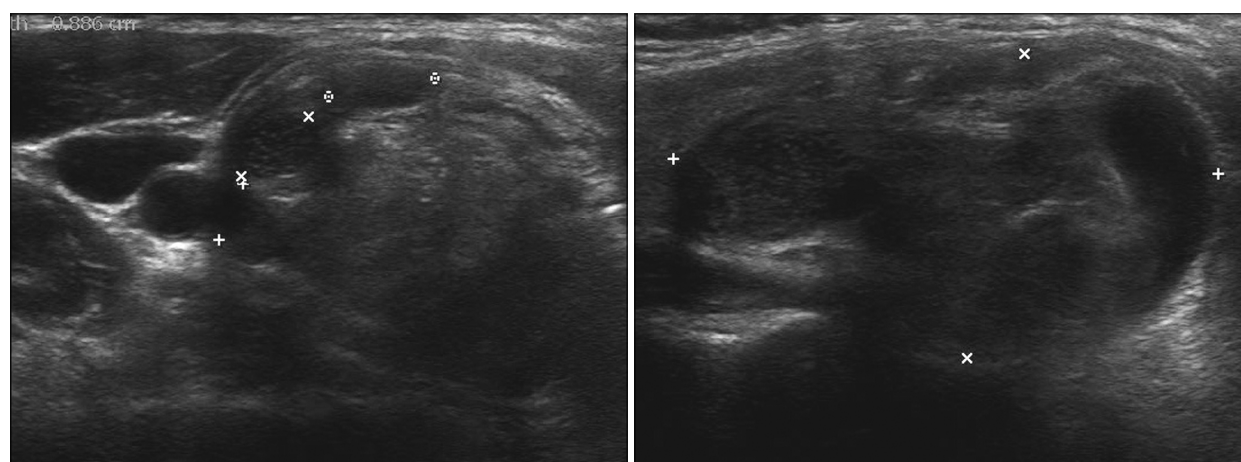


Fig. 1. Ultrasonographic scans showing right lobe was mostly occupied with a solid nodule of 44×23×25 mm with markedly inhomogeneous hypoechogenicity and hyperechoic lesion.

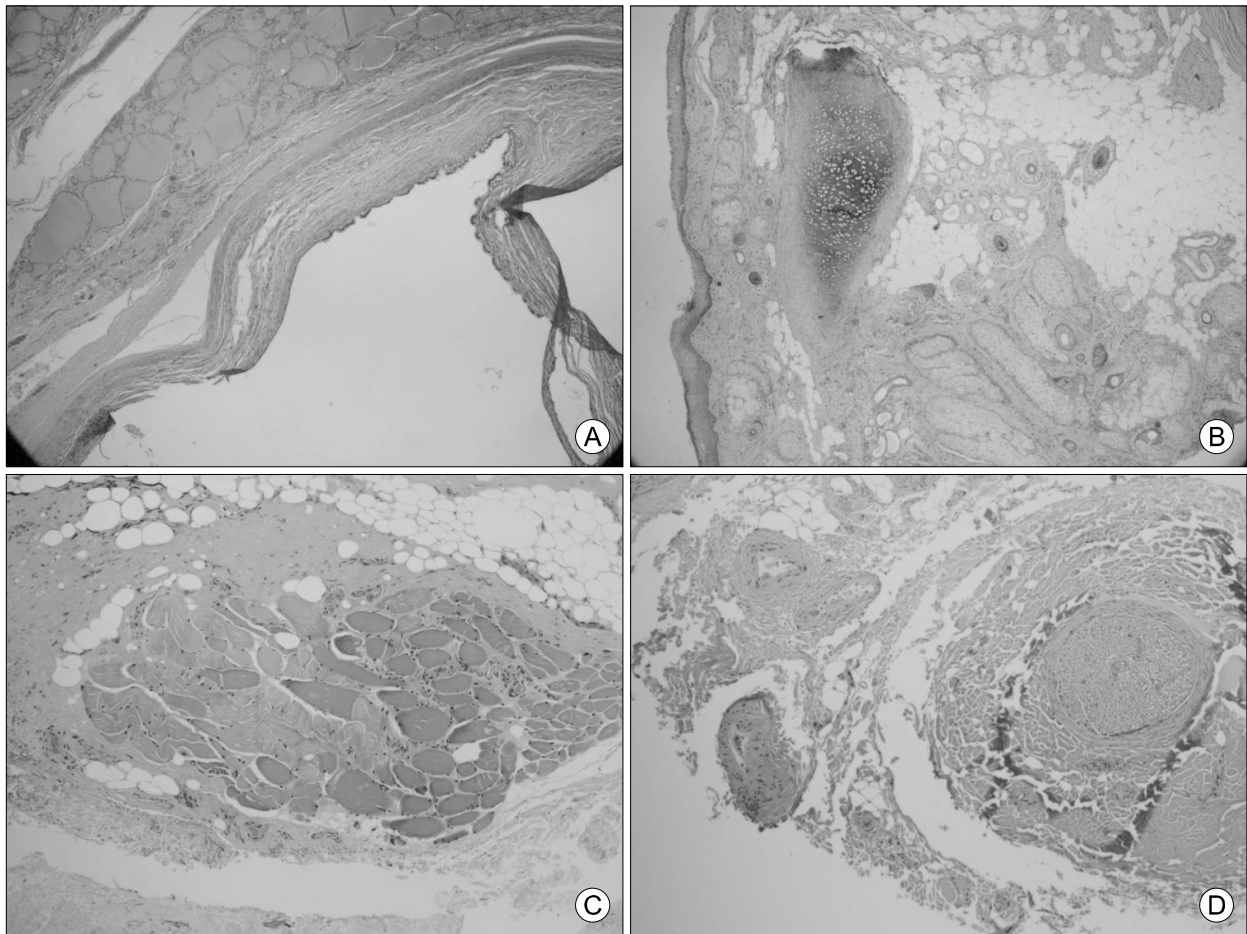


Fig. 3. Histologic findings of the benign thyroid teratoma. (A) Squamous cell epithelium and many skin appendages including hair follicles, hairs and many sebaceous glands and sweat glands are seen (H&E staining, $\times 50$). (B) The cystic wall partly contains cartilage and bony tissue (H&E staining, $\times 50$). (C) Mature fat tissue and smooth muscle are observed (H&E staining, $\times 100$). (D) Nerves and vessel structures are also seen (H&E staining, $\times 100$).

structures with mucin. As there was no infiltration into surrounding tissues, or metastasis to lymph nodes in the permanent pathology, possibility of a malignant teratoma was ruled out. Microscopic examination revealed benign mature cystic teratoma (with cartilage, bone and fat tissue, and skin appendages with hairs) (Fig. 3) and there was no abnormal finding in both ovary by abdomen and pelvic US.

After lobectomy, patient took 50 mcg of levothyroxine sodium and thyroid function remains normal and no signs of recurrence have been found for 8 years.

Discussion

Teratomas are germ cell tumors featuring tissues from the three primordial layers (i.e., mesoderm, endo-

derm, and ectoderm). A subset of these tumors is found in the neck, and a few of these are located within or in close anatomic relation to the thyroid. However, a thyroid teratoma is a very rare form of germ cell tumor.⁷⁾ Although there is controversy about diagnostic criteria of thyroid teratoma, in general, cervical tumor is regarded as thyroid teratoma when it meets one or more of these criteria: 1) tumor occupies part of the thyroid, 2) tumor has direct continuity with thyroid gland, 3) the thyroid is not completely developed, but teratoma exists in the neck.^{3,5,8-10)} With this criteria, the tumor in this case met two criteria by occupying part of the thyroid and being located in direct continuity with the thyroid. And histological result also showed that the tumor contains all three germ layers components.

In this case, an adolescent girl came with neck enlargement and discomfort. Palpated mass was rapidly enlarged within 2 months, and she complained about compression symptoms by it. Our patient's neck pain and discomfort might be caused by increase of fluid in cystic part of the teratoma. Several references suggest that irritation due to increased secretion from pancreatic tissue in mature teratoma is thought to play a role in cystic formation and inflammatory reactions.^{11,12)} However, in our patient's histologic result, pancreatic tissue was not found.

In US, the tumor has well defined anterior margin and hypoechogenicity. However, the hyperechoic internal lesions within the nodule made it difficult to differentiate thyroid cancer from benign thyroid nodule. The posterior margin of the tumor was not clear due to excessive attenuation of US just as teratoma with fatty tissue and hair.¹³⁾ Schmitz et al.¹³⁾ reported mixed hyperechoic, hypoechoic, and nonechoic lesions as characteristics of teratoma in US evaluation. However, they also added that teratoma-specific US features are not clear.

Although FNAC is considered as the standard for preoperative evaluation of thyroid nodules, the value of this has been questioned for thyroid nodules 4 cm or larger. According to literatures, the false negative rates of FNAC in thyroid nodules 4 cm or larger are 2% to 10%. Among patients with non-diagnostic FNAC, the risk of malignancy was 27%.¹⁴⁻¹⁷⁾ Moreover, incidence of a clinically significant thyroid cancer within the ≥ 4 cm nodule was 22%.¹⁸⁾

Because the result of FNAC in our case was cystic liquid and non-diagnostic, we could not completely rule out the possibility of malignancy. Additionally, our patient was younger than 18 years old and studies suggest that children show higher rate of cancer more than adult.¹⁹⁾ Therefore, we decided to perform surgical treatment, and the result of permanent biopsy revealed mature cystic thyroid teratoma. After we get final diagnosis we could retrace our differential diagnosis and we rediscover that the tumor was suspicious for teratoma. This case showed another rationality for surgical resection in thyroid nodule larger than 4 cm in adolescence.

When thyroid teratoma is suspected, further evaluation is essential for ruling out other diagnosis including lymphoid hyperplasia, lymphoma, cystic hygroma, and thyroglossal duct cyst.²⁾ If fatty components or calcifications are found in imaging tests, further aggressive testing should be performed to differentiate malignant disease.¹⁹⁾ Approximately 10% of thyroid teratomas are found after puberty, and less than half of cases are proven benign according to the literature.^{4,11)} Development of benign teratoma can be influenced by several triggers and can be accelerated in adolescents and adults. Majority of neonatal and pediatric teratomas display benign histology, however they have significant morbidity because of respiratory distress or the vital structures malformation in the neck, whereas a large proportion of those appearing in adult life are malignant.²⁰⁾ Diagnostic imaging tests are not enough to differentiate malignant tumors from benign tumors, so a surgical resection is needed.

Until now there was only a single case of benign thyroid teratoma reported in South Korea and patient was 54-year-old woman.³⁾ Therefore this case would be the first case of benign thyroid teratoma of adolescent, reported in South Korea.

In summary, the patient had an intrathyroidal mass larger than 4 cm with a clear boundary showing a combination of hypoechoic and hyperechoic lesions and the result of FNA was non-diagnostic. As patient was younger than 18 years old and her mass increased rapidly, we performed lobectomy and benign teratoma was diagnosed. When large thyroid nodule is detected in adolescence and malignancy could not be ruled out, final diagnosis should be made with surgical resection.

Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

References

- 1) Fan SQ, Liang QC, Jiang Y. *Thyroid teratoma in an 11-month-old infant. Int J Surg* 2008;6(6):462-4.

- 2) Kim JY, Cho YZ, Lee KW, Lim DM, Park KY, Kim BJ. *A case of adrenal teratoma. Endocrinol Metab* 2011;26(3):272-5.
- 3) Oak CY, Kim HK, Yoon TM, Lim SC, Park HB, Park HC, *et al. Benign teratoma of the thyroid gland. Endocrinol Metab (Seoul)* 2013;28(2):144-8.
- 4) Nishihara E, Miyauchi A, Hirokawa M, Kudo T, Ohye H, Ito M, *et al. Benign thyroid teratomas manifest painful cystic and solid composite nodules: three case reports and a review of the literature. Endocrine* 2006;30(2):231-6.
- 5) Oka K, Okane M, Okuno S, Kawasaki T, Yonekawa N, Okano M, *et al. Congenital cervical immature teratoma arising in the left lobe of the thyroid gland. APMIS* 2007;115(1):75-9.
- 6) Barksdale EM Jr, Obokhare I. *Teratomas in infants and children. Curr Opin Pediatr* 2009;21(3):344-9.
- 7) Kimler SC, Muth WF. *Primary malignant teratoma of the thyroid: case report and literature review of cervical teratomas in adults. Cancer* 1978;42(1):311-7.
- 8) Thompson LD, Rosai J, Heffess CS. *Primary thyroid teratomas: a clinicopathologic study of 30 cases. Cancer* 2000; 88(5):1149-58.
- 9) Saphir O. *Teratoma of the neck. Am J Pathol* 1929;5(3):313-22 3.
- 10) Bale GF. *Teratoma of the neck in the region of the thyroid gland; a review of the literature and report of 4 cases. Am J Pathol* 1950;26(4):565-79.
- 11) Sommerlad BC, Cleland WP, Yong NK. *Physiological activity in mediastinal teratomata. Thorax* 1975;30(5):510-5.
- 12) Kallis P, Treasure T, Holmes SJ, Griffiths M. *Exocrine pancreatic function in mediastinal teratomata: an aid to preoperative diagnosis? Ann Thorac Surg* 1992;54(4):741-3.
- 13) Schmitz H, Spelsberg F, Hefele-Roedel B, Meister P. *Ultrasound diagnosis of benign teratoma of the thyroid gland. Bildgebung* 1990;57(3-4):93-5.
- 14) Pinchot SN, Al-Wagih H, Schaefer S, Sippel R, Chen H. *Accuracy of fine-needle aspiration biopsy for predicting neoplasm or carcinoma in thyroid nodules 4 cm or larger. Arch Surg* 2009;144(7):649-55.
- 15) McCoy KL, Jabbour N, Ogilvie JB, Ohori NP, Carty SE, Yim JH. *The incidence of cancer and rate of false-negative cytology in thyroid nodules greater than or equal to 4 cm in size. Surgery* 2007;142(6):837-44; discussion 44 e1-3.
- 16) Yoon JH, Kwak JY, Moon HJ, Kim MJ, Kim EK. *The diagnostic accuracy of ultrasound-guided fine-needle aspiration biopsy and the sonographic differences between benign and malignant thyroid nodules 3 cm or larger. Thyroid* 2011; 21(9):993-1000.
- 17) Kuru B, Gulcelik NE, Gulcelik MA, Dincer H. *The false-negative rate of fine-needle aspiration cytology for diagnosing thyroid carcinoma in thyroid nodules. Langenbecks Arch Surg* 2010;395(2):127-32.
- 18) Wharry LI, McCoy KL, Stang MT, Armstrong MJ, LeBeau SO, Tublin ME, *et al. Thyroid nodules (>=4 cm): can ultrasound and cytology reliably exclude cancer? World J Surg* 2014;38(3):614-21.
- 19) Gupta A, Ly S, Castroneves LA, Frates MC, Benson CB, Feldman HA, *et al. A standardized assessment of thyroid nodules in children confirms higher cancer prevalence than in adults. J Clin Endocrinol Metab* 2013;98(8):3238-45.
- 20) Colton JJ, Batsakis JG, Work WP. *Teratomas of the neck in adults. Arch Otolaryngol* 1978;104(5):271-2.