

Recurrence of papillary thyroid carcinoma from the residual pyramidal lobe: a case report and literature review

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Abstract

Rationale: Recurrence of papillary thyroid carcinoma (PTC) usually requires a second operation, which carries a high complication rate, especially if central neck dissection (CND) is necessary. Recurrent PTC from pyramidal lobe is a rare entity, which is mainly due to non-standardized operation. However, literature on this topic is limited.

Patients concerns: Here, we present a case of a 46-year-old woman with the recurrence of PTC from the thyroid pyramidal lobe (PL) following two thyroid operations.

Diagnoses: The final pathological result revealed recurrent PTC from the residual pyramidal lobe tissue.

Interventions: The resection of the residual PL, the pretracheal nodes and the Delphian nodes plus intraoperative neuromonitoring (IONM) were performed, followed by TSH suppression therapy.

Outcomes: After two previous operations, the recurrent PTC from the residual pyramidal lobe tissue was completely resected. Until the latest follow-up, the patient had an excellent response to the third intervention.

Lessons: Complete excision of the PL is mandatory in patients with PTC if thyroid surgery was indicated, considering the potential risk of recurrence from the PL and the high incidence of multifocality of PTC.

Abbreviations: CLND = central lymph node dissection, ETE = extrathyroidal extension, FNA = fine-needle aspiration cytology, IONM = intraoperative neuromonitoring, MRND = modified radical neck dissection, NG = nodular goiter, PL = pyramidal lobe, PTC = papillary thyroid carcinoma, RAI = radioactive iodine, UPTC = upper neck papillary thyroid carcinoma, US = ultrasound.

Keywords: papillary thyroid carcinoma, pyramidal lobe, recurrence

1. Introduction

The detection of thyroid carcinoma has been increasing in recent years due to the widespread utility of the high-resolution ultrasonography (US) and US-guided fine-needle aspiration cytology (FNA). Papillary thyroid carcinoma (PTC) is the most common histologic type of differentiated thyroid cancer, accounting for about 80% of thyroid carcinoma, and it is

characterized by early regional lymph metastasis, but has a favorable prognosis with the 10-year relative survival rate of 93%.^[1] Although PTC grows slowly, differentiates well, and has an indolent clinical course, the recurrence is not uncommon in patients with PTC, with a recurrence rate of 5% to 21%.^[2,3] Tumor size, extrathyroidal extension (ETE), multifocality, age, extranodal spread, lymph node ratio, and male sex were reported to have a predictive value for the recurrence of PTC.^[4-6] It was reported that the central compartment lymph nodes and proper thyroid tissue of the thyroid bed were the most common recurrence sites following thyroidectomy.^[1] The carcinoma recurrence from the pyramidal lobe (PL) of thyroid is quite rare, with quite a few cases recorded in the literatures. Here, we present a case of a 46-year-old woman with the recurrence of PTC from the pyramidal lobe following two thyroid operations. Informed written consent was obtained from the patient for publication of this case report and accompanying images, which was approved by the ethics committee of West China Hospital, Sichuan University.

2. Case presentation

A 46-year-old woman sought medical attention due to a nodule located in the right thyroid lobe, which was found incidentally in physical examination 7 years ago (2011.08). Subsequently, she was referred to a regional hospital. An ultrasound examination revealed a 36×24×18mm solid hypoechoic nodule located within the upper pole of the right thyroid lobe, and TSH, FT3,

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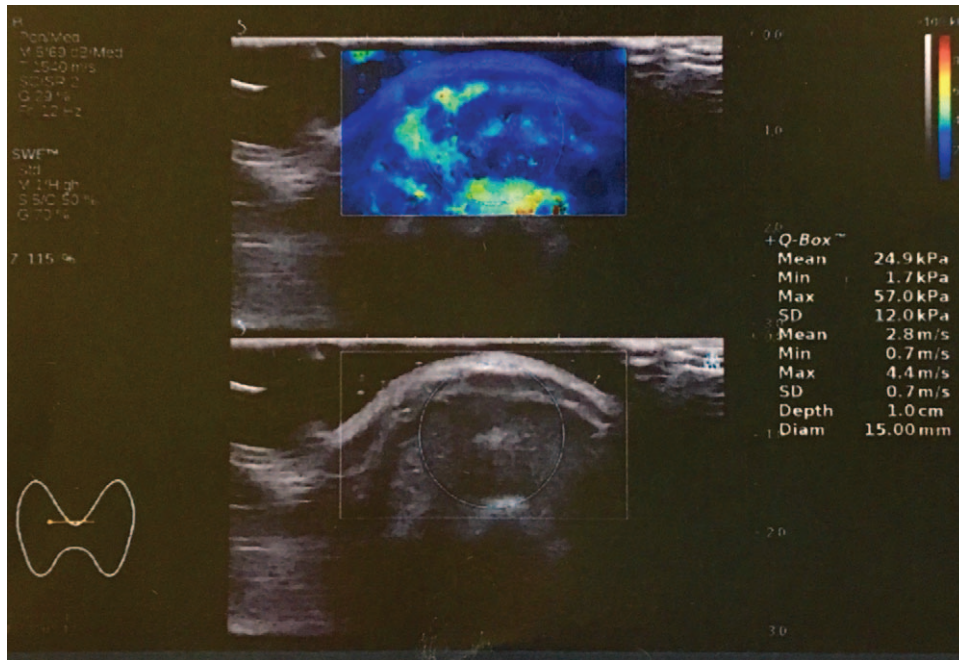


Figure 1. Ultrasound detected a solid hypoechoic and irregular shape nodule in size of 21×10×16mm above the thyroid cartilage (transverse view).

and FT4 were all in the normal ranges. After the diagnosis of PTC was confirmed by the US-guided FNA, right lobectomy plus isthmusectomy was performed based on the preoperative evaluation of the recurrence risk and the intraoperative findings in her regional hospital. A daily oral supplement with a dosage of 75 μg levothyroxine was indicated following surgery.

After a follow-up of 44 months (2015.04) following the initial surgery, a hyperechoic solid nodule in the left lobe and abnormal lymph nodes (10×5 mm) in the right central compartment were

detected by US. The right central neck recurrent PTC and the diagnosis of nodular goiter were considered. After confirmed by the US-guided FNA, the completion thyroidectomy plus right central neck dissection was performed in the same regional hospital. This patient received 2 radioactive iodine treatments 1 and 6 months after the second surgery respectively (160 mCi and 180 mCi respectively). The post-treatment whole-body ¹³¹I imaging showed no significantly increased accumulation of ¹³¹I. The suppressive therapy with a dosage of 100 μg levothyroxine

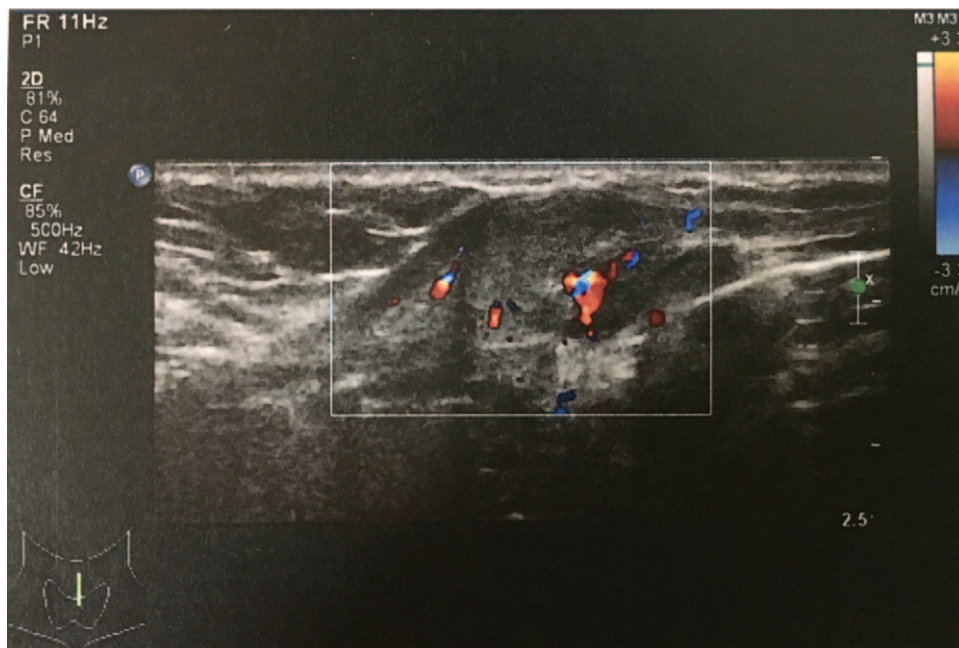


Figure 2. Ultrasound detected a solid hypoechoic and irregular shape nodule in size of 21×10×16mm above the thyroid cartilage (longitudinal view).

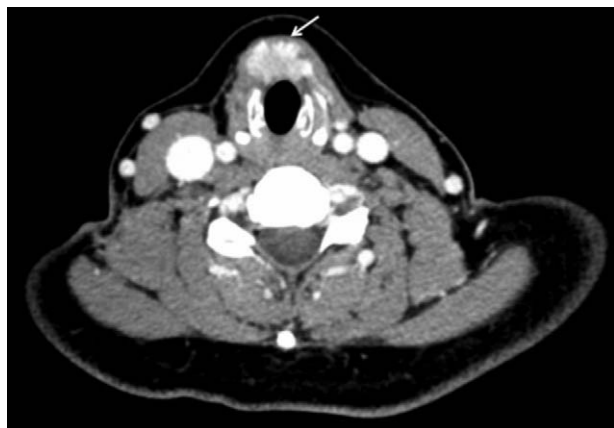


Figure 3. An obvious enhancing nodular lesion above the thyroid cartilage was detected by enhanced CT scan (white arrow).

was administered to this patient after the second surgery. Follow-up surveillance including neck ultrasound imaging, thyroid function measurement, as well as marker analysis of thyroglobulin (Tg) and TgAb (Tg-antibody) was planned every 6 months.

Twenty-seven months after the second operation (2017.07), a 21×10×16 mm solid hypoechoic nodule with irregular shape was detected above the thyroid cartilage during ultrasonography in our hospital (Figs. 1 and 2). And the neck enhanced CT showed an obvious enhancing nodular lesion above the thyroid cartilage (Fig. 3). TSH: 0.012 mU/L, FT3: 5.00 pmol/L, FT4: 24.22 pmol/L, Tg: 73.18 ug/L, TgAb: 10.14 IU/mL. The recurrence of PTC was suspected, which was confirmed by FNA. The resection of the residual PL, the pretracheal nodes and the Delphian nodes plus IONM were performed. Final histopathological examination confirmed PTC arising from the pyramidal lobe. A daily oral supplement with a dosage of 100 µg levothyroxine was indicated following surgery. The patient was disease free until the latest follow-up (2018.08). The treatment history of this patient was summarized in Table 1.

3. Discussion and lessons

Pyramidal lobe is an embryological remnant of thyroglossal duct and is considered a normal compartment of thyroid gland.^[7] The incidence of PL varies from 15% to 75% in anatomical studies,^[7] and the shape, position, appearance as well as size vary greatly in individuals. Normally, thyroid tissue in pyramidal lobe is usually not active, hence is not visible on scintigraphy.^[8] Complete excision of the PL is mandatory in patients with PTC considering the high incidence of multifocality of PTC; Furthermore, the function of the residual PL will be activated when it is not

removed during indicated total thyroidectomy. Both of the above-mentioned factors make the PL a potential place of recurrent PTC. PTC arising from the PL is quite rare, and the recurrence of PTC from the pyramidal lobe is mainly due to the irregular operation. Once the diagnosis of recurrent PTC of the PL is established, reoperation is the first choice if possible.

Previous studies have emphasized the importance of pyramidal lobe in thyroid surgery.^[7-12] However, only a single report described a South Korean patient with recurrent PTC in the residual pyramidal lobe tissue. In Lee study, a right modified radical neck dissection with regional dissection around the remnant pyramidal lobe was performed for the patient with recurrence of PTC in a remnant PL.^[11] The treatment principle is in line with the NCCN guideline in our case,^[1] where the resection of the residual PL, the pretracheal nodes and the Delphian nodes plus IONM were performed. Sporadic cases of primary (instead of recurrent) PTC from the pyramidal lobe were reported.^[13-16] Yoon et al enrolled 1107 patients with PTC in his study, finding the prevalence of occult malignancy in surgically resected pyramidal lobe tissue was 3.56%.^[13] Ha et al reported a 48-year-old woman with primary PTMC in the PL and both thyroid lobes, and total thyroidectomy and central compartment neck dissection were performed for this patient.^[16] Santrac et al enrolled 753 patients with PTC from 2003 to 2013 in his institution, but only 3 patients (0.4%) suffered from solitary PTC primarily from the PL. He concluded that the appearance of primary PTC focus in the PL was rare, the lesions could be easily diagnosed due to the accessibility to palpation and FNA, and emphasized the extent of the surgery for such patients should be discussed by experienced surgeons.^[15] The English literature regarding (primary or recurrent) PTC from the pyramidal lobe, which were electronically searched from Embase, Medline, and PubMed up to and including October 2018 were summarized in Table 2. Due to a paucity of literature on this topic, the actual incidence of PTC in the pyramidal lobe is unclear. Anyway, it is important to keep in mind that careful preoperative evaluation and standardized removal of the pyramidal lobe during thyroid surgery is essential.

On the other hand, there is a lack of knowledge of the recurrent PTC from PL. Therefore, some differential diagnoses should be emphasized. Thyroglossal duct cyst and pyramidal lobe have the same origin, and the similar anatomical position; furthermore, the incidence of diseases from PL is lower than that of thyroglossal duct cyst. Thus, PTC from PL is easily misdiagnosed as thyroglossal duct cyst. Thyroglossal duct cyst usually presents rounded, liquid echo free zone on ultrasound, and there is no blood flow signal inside the lesion. The presence of features consistent with PTC, such as irregular margin, hypoechoic, microcalcifications, and taller than wide shape, on ultrasound should raise suspicion for malignancy. The PL is intrinsically dense on CT, with attenuation and enhancement identical to the main thyroid gland on non-enhanced and contrast enhanced CT

Table 1

Treatment history.

Date	Final diagnosis	Surgical procedure	Postoperative treatment
2011.08	PTC within the right lobe	Right lobectomy plus isthmusectomy	TSH suppression therapy
2015.04	NG+ right central neck recurrent PTC	Completion thyroidectomy plus right central neck dissection	TSH suppression therapy + RAI
2017.07	Recurrent PTC from the PL	Resection of the residual PL, the pretracheal nodes and the Delphian nodes plus IONM	TSH suppression therapy

PTC=papillary thyroid carcinoma, NG=nodular goiter, PL=pyramidal lobe, IONM=intraoperative nerve monitoring, RAI=radioactive iodine.

Table 2**Review of published reports regarding PTC from the pyramidal lobe.**

Author	Year	Country	No. of cases	Primary or recurrent PTC	Surgical procedure	Lessons
Lee, Y.S. ^[11]	2011	Korea	1	Recurrent	A right modified radical neck dissection and regional dissection around the remnant pyramidal lobe	Complete surgical resection of the pyramidal lobe is mandatory for treatment of PTC.
Ha, T.K. ^[16]	2014	Korea	1	Primary	Total thyroidectomy and central compartment neck dissection	Preoperative evaluation of the PL by US should be included considering possible multifocality of PTC.
Santrac, N. ^[15]	2014	Serbia	3	Primary	Total thyroidectomy and central neck dissection (uni- or bilateral), with sentinel lymph node biopsy in level III/IV of both jugulo-carotid regions	PTC within PL is very rare and only individual experiences can be discussed regarding the extent of the surgery.
Zizic, M. ^[14]	2016	USA	8	Primary	Total thyroidectomy with nodal dissection	UPTC represents a clinical diagnosis that consists of a heterogeneous group, and the correct pathologic subcategory diagnosis after initial surgery in a UPTC patient allows for optimal management.
Yoon, S.G. ^[13]	2017	Korea	39	Primary	Total thyroidectomy/lobectomy with pyramidal lobe resection with CLND/MRND	PL-dominant PTC is rare; however, it is related to the presence of poor prognostic factors. Proper thyroid resection with regional lymph node excision should be performed for these patients.
Current study	2018	China	1	Recurrent	Resection of the residual PL, the pretracheal nodes and the Delphian nodes plus IONM	Standardized surgical procedure (complete removal of the pyramidal lobe) is mandatory in indicated thyroidectomy to facilitate postoperative radioiodine therapy (for total thyroidectomy) and remove the potential origin of recurrent PTC.

PTC = papillary thyroid carcinoma, PL = pyramidal lobe, CLND = central lymph node dissection, MRND = modified radical neck dissection, IONM = intraoperative nerve monitoring, US = ultrasound, UPTC = upper neck papillary thyroid carcinoma.

images respectively, while thyroglossal duct cysts are thin-walled, smooth, well-defined homogeneously water-density lesions. Furthermore, radionuclide imaging can also be performed to differentiate diseases. Other differential diagnoses include PTC from ectopic thyroid tissue in the middle line of the neck, Delphian node metastasis, bronchogenic cysts and so on. Finally, US-guided FNA is a minimally invasive and safe procedure that can give a definite diagnosis.

In summary, standardized surgical procedure (complete removal of the pyramidal lobe) is mandatory in indicated thyroidectomy, which can not only facilitate postoperative radioiodine therapy (for total thyroidectomy) but also can remove the potential origin of recurrent PTC.

Author contributions

Conceived and designed the study: Mingjun Wang, Xiuhe Zou, Zhihui Li, Jingqiang Zhu.

Performed the experiments: Mingjun Wang, Xiuhe Zou.

Wrote the paper: Mingjun Wang.

Final approval of the version to be published: Jingqiang Zhu.

Investigation: Xiuhe Zou.

Resources: Xiuhe Zou, Zhihui Li.

Supervision: Jingqiang Zhu.

Validation: Zhihui Li.

Writing – original draft: Mingjun Wang.

Writing – review & editing: Jingqiang Zhu.

References

- [1] NCCN Clinical Practice Guidelines in oncology. Thyroid carcinoma. Version 2. 2015. http://www.nccn.org/professionals/physician_gls/pdf/thyroid.pdf.
- [2] Grant CS. Recurrence of papillary thyroid cancer after optimized surgery. *Gland Surg* 2015;4:52–62.
- [3] Liu FH, Kuo SF, Hsueh C, et al. Postoperative recurrence of papillary thyroid carcinoma with lymph node metastasis. *J Surg Oncol* 2015;112:149–54.
- [4] Nam SH, Roh JL, Gong G, et al. Nodal factors predictive of recurrence after thyroidectomy and neck dissection for papillary thyroid carcinoma. *Thyroid* 2018;28:88–95.
- [5] Chereau N, Buffet C, Tresallet C, et al. Recurrence of papillary thyroid carcinoma with lateral cervical node metastases: predictive factors and operative management. *Surgery* 2016;159:755–62.
- [6] Siddiqui S, White MG, Antic T, et al. Clinical and pathologic predictors of lymph node metastasis and recurrence in papillary thyroid microcarcinoma. *Thyroid* 2016;26:807–15.
- [7] Geraci G, Pisello F, Li Volsi F, et al. The importance of pyramidal lobe in thyroid surgery. *G Chir* 2008;29:479–82.
- [8] Milojevic B, Tosevski J, Milisavljevic M, et al. Pyramidal lobe of the human thyroid gland: an anatomical study with clinical implications. *Rom J Morphol Embryol* 2013;54:285–9.
- [9] Sinos G, Sakorafas GH. Pyramidal lobe of the thyroid: anatomical considerations of importance in thyroid cancer surgery. *Oncol Res Treat* 2015;38:309–10.
- [10] Zivic R, Radovanovic D, Vekic B, et al. Surgical anatomy of the pyramidal lobe and its significance in thyroid surgery. *S Afr J Surg* 2011;49:110112, 114 passim.
- [11] Lee YS, Kim KJ, Kim BW, et al. Recurrence of papillary thyroid carcinoma in a remnant pyramidal lobe. *ANZ J Surg* 2011;81:304.
- [12] Braun EM, Windisch G, Wolf G, et al. The pyramidal lobe: clinical anatomy and its importance in thyroid surgery. *Surg Radiol Anat* 2007;29:21–7.
- [13] Yoon SG, Yi JW, Seong CY, et al. Clinical characteristics of papillary thyroid carcinoma arising from the pyramidal lobe. *Ann Surg Treat Res* 2017;92:123–8.
- [14] Zizic M, Faquin W, Stephen AE, et al. Upper neck papillary thyroid cancer (UPTC): a new proposed term for the composite of thyroglossal duct cyst-associated papillary thyroid cancer, pyramidal lobe papillary thyroid cancer, and Delphian node papillary thyroid cancer metastasis. *Laryngoscope* 2016;126:1709–14.
- [15] Santrac N, Besic N, Buta M, et al. Lymphatic drainage, regional metastases and surgical management of papillary thyroid carcinoma arising in pyramidal lobe—a single institution experience. *Endocr J* 2014;61:55–9.
- [16] Ha TK, Kim DW, Park HK, et al. Papillary thyroid microcarcinoma in a thyroid pyramidal lobe. *Ultrasonography* 2014;33:303–6.