

Primary Thyroid Squamous Carcinoma: A Case Report and Related Literature Review

Min Liu¹, Yanqiu Song², Tao Han¹, Liang Guo³, Bailong Liu^{1*} and Lihua Dong^{1*}

¹Department of Radiation Oncology, The First Hospital, Jilin University, 71 Xinmin Street, Changchun, 130021, Peoples Republic of China

²Cancer Center, The First Hospital, Jilin University, 71 Xinmin Street, Changchun, 130021, Peoples Republic of China

³Department of Pathology, The First Hospital, Jilin University, 71 Xinmin Street, Changchun, 130021, Peoples Republic of China

*Corresponding authors: Bailong Liu, Department of Radiation Oncology, the First Hospital, Jilin University, 71 Xinmin Street, Changchun, 130021, Peoples Republic of China, Tel: 8643188782468; E-mail: bailong3385@163.com

Lihua Dong, Department of Radiation Oncology, the First Hospital, Jilin University, 71 Xinmin Street, Changchun, 130021, Peoples Republic of China, Tel: 86-431-88782468; E-mail: lijie200461@126.com

Received date: September 01, 2014, Accepted date: October 19, 2014, Published date: October 27, 2014

Copyright: © 2014 Liu M, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Abstract

Primary Squamous Carcinoma of Thyroid (PSCT) is a rare but distinct clinicopathological disease. Due to the paucity, optimal intervention strategy has not been established yet. PSCT was once viewed as radioresistant. Here we present a rare case of PSCT in which postoperative radiotherapy was effective for local control and maintenance of progression free survival (PFS) of more than 10 months after R2 resection. Our case will help to identify the role of adjuvant radiotherapy in the treatment of PSCT and extend our understanding of this rare thyroid carcinoma.

Keywords: Primary thyroid squamous carcinoma; Radiotherapy

Introduction

PSCT is a distinct disease seldom encountered in clinical practice. Generally, it has an aggressive behavior with a median survival of 9.0 months [1]. Up to date, only two meta-analyses have been conducted [1,2] and all the other reports were described in case reports or series. Because of the low incidence, best treatment options and related prognostic factors are still underdetermined. Here, we present a case of PSCT in which R2 resection followed by adjuvant radiotherapy attained a favorable PFS of at least 10 months.

Case Report

A 63-year-old man presented in October, 2013 with a history of anterior neck mass for 4 months and hoarseness for 2 months. On physical examination, a regular 2.5 cm × 2.0 cm and 5.0 cm × 3.0 cm mass was palpated in the right and left lobe of thyroid respectively without tenderness, moving up and down with swallowing. Trachea shifted to the right. No lymphadenopathy was revealed in bilateral neck. His past history was unremarkable.

Laryngoscopy showed left vocal cord paralysis. The following ultrasound of the neck demonstrated multiple thyroid nodules scattered in bilateral lobes, the largest measuring about 64 × 49 mm located in the left lobe, occupying almost the entire left lobe with calcification. A 24 × 18 mm regular nodule was located in the right lobe with clear border. The neck CT showed a huge lesion in the left lobe of thyroid with significant calcification compressing the trachea to the left and a low-density mass located in the right lobe without calcification. The level of CEA was normal. Whole-body bone scan revealed no abnormal radionuclide uptake. Thoracic CT and esophageal barium meal image were unremarkable.

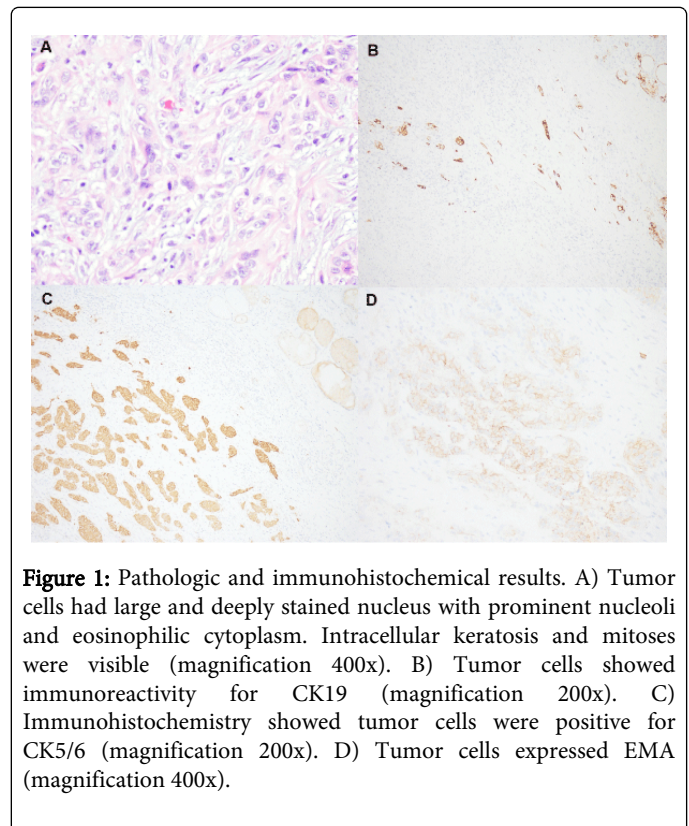


Figure 1: Pathologic and immunohistochemical results. A) Tumor cells had large and deeply stained nucleus with prominent nucleoli and eosinophilic cytoplasm. Intracellular keratinization and mitoses were visible (magnification 400x). B) Tumor cells showed immunoreactivity for CK19 (magnification 200x). C) Immunohistochemistry showed tumor cells were positive for CK5/6 (magnification 200x). D) Tumor cells expressed EMA (magnification 400x).

On November 19th, 2013, surgery was performed during which a tough 5.0 cm × 6.5 cm × 5.5 cm mass was found in the left lobe of thyroid with severe adhesion of nearby normal tissues, unable to be resected completely. Thus, the entire nodule in the right lobe, part of the mass in the left and thyroid gland were excised. The definite

pathologic results showed as followings: the lesion of left lobe was thyroid squamous carcinoma (Figure 1A) and the lesion of right lobe was nodular goiter. The postoperative immunochemistry of the lesion of left lobe showed: CK19(+) (Figure 1B), CK5/6(+) (Figure 1C), EMA(+) (Figure 1D), P63(+), P53(+90%), Calcitonin(), TG(-), CgA(-). Based on the above, PSCT with a stage of T4N0M0 was diagnosed.

disease (SD) response (Figure 2C). The patient remains well and PFS has reached 10 months now.

Discussion

PSCT is exceedingly rare, with an incidence of 0.7% of all the thyroid carcinomas [3]. PSCT usually occurs in the elderly and shows a predilection of female [1]. The most common symptoms include rapidly enlarged mass of anterior neck, hoarseness, dysphagia and dyspnea resulted from compression of adjacent normal organs such as recurrent laryngeal nerve, oesophagus and trachea. Due to the high malignancy, only a small proportion of patients, 22.5% by Cho JK et al. had lesion confined to thyroid gland [1]. Bone was the most common site of distant metastasis [1]. Rare metastasis such as heart has also been documented [4]. PSCT bears a grim prognosis with shortest reported survival of 17 days [5].

The diagnosis of PSCT is challenging, for the possibility of metastasis of squamous cell carcinoma (SCC) of other sites, especially the upper aerodigestive tract, and invasion of SCC nearby should be fully ruled out. Yucel H et al. reported a case of PSCT with a history of radioactive iodine treatment for hyperthyroidism 25 years ago [6]. However, our patient was previously healthy, without any history of hyperthyroidism or treatment of radioactive iodine. The final diagnosis of PSCT should be made after the comprehensive consideration based on the combination of clinical, radiographic, pathologic and immunohistochemical results. Radiologically, PSCT is lack of specific findings [2,7]. Immunohistochemistry is essential for differential diagnosis. PSCT often express CK19 [3,5] while a negative calcitonin and thyroglobulin is useful for excluding the diagnosis of medullary thyroid carcinoma and malignancy arising from thyroid follicular cells respectively [5]. In this patient, CK19(+), Calcitonin() and Thyroglobulin() are critical for the final diagnosis of PSCT. PSCT also can possess BRAF mutation [8].

Recently, in the multivariate analysis by Cho JK et al. [1], R0 resection was considered as the only factor associated with improved survival. Unfortunately, the majority presents in an advanced stage, making the radical resection impossible. How to improve the treatment outcomes of cases that are unresectable or cannot be removed completely? Modalities besides surgery need to be further clarified. PSCT was once considered as radioresistant [9]. Tunio MA et al. documented an unresectable PSCT in which external beam radiotherapy was ineffective [10]. Our case verified the efficacy of postoperative radiotherapy for a patient with residual tumor. Although postirradiation CT scan didn't demonstrate significant shrinkage of tumor, discomfort of the patient relieved greatly. The patient achieved a good local control with favorable PFS of at least 10 months. Aggressive local excision and adjuvant radiotherapy contributed to the long-term PFS of 21 months reported by Shrestha M et al. [11] Palliative radiotherapy had been proved to be effective in relieving shortness of breath and pain and guaranteed a 1-year survival of good quality in an unresectable patient of PSCT [12]. Chemotherapy usually fails to work in PSCT [9].

In conclusion, PSCT was rare usually progressing rapidly and bearing a poor prognosis. Radical surgical intervention plus adjuvant radiotherapy might contribute to the extended PFS. Adjuvant radiotherapy is effective in local control for postoperative PSCT patients with residual tumor.

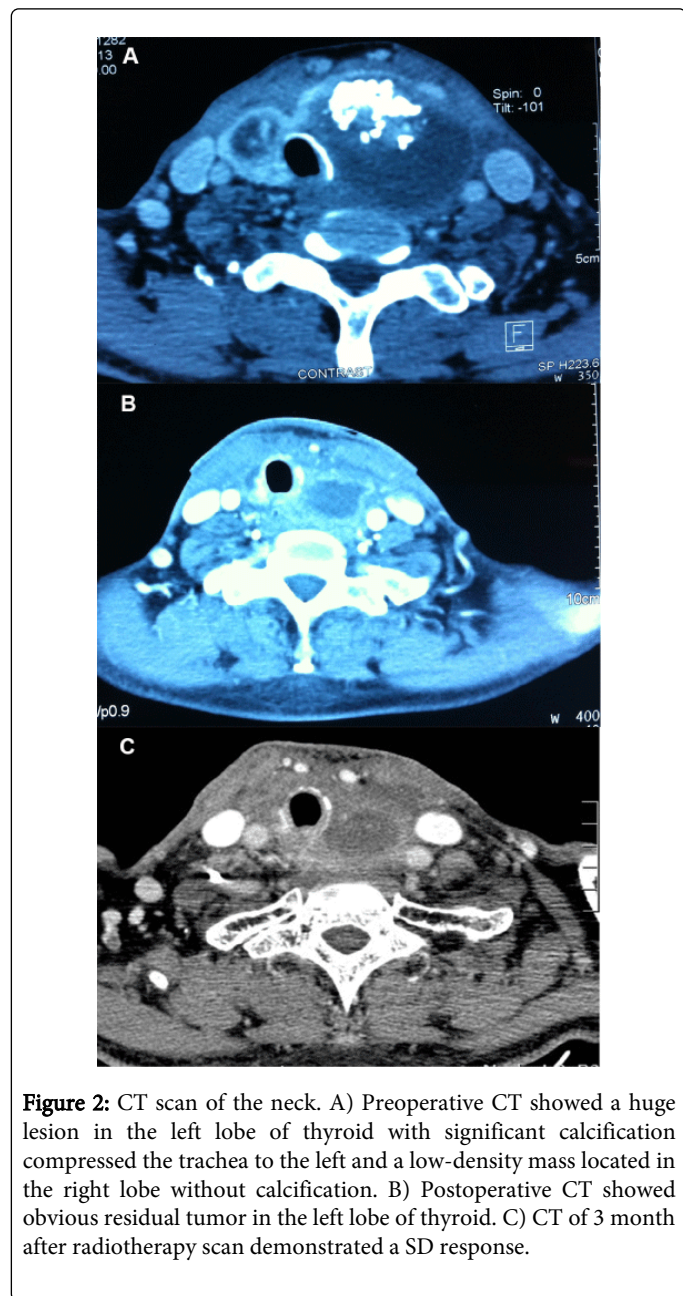


Figure 2: CT scan of the neck. A) Preoperative CT showed a huge lesion in the left lobe of thyroid with significant calcification compressed the trachea to the left and a low-density mass located in the right lobe without calcification. B) Postoperative CT showed obvious residual tumor in the left lobe of thyroid. C) CT of 3 month after radiotherapy scan demonstrated a SD response.

Then the patient recovered uneventfully and hoarseness relieved remarkably. One month after surgery, comparing with CT before operation (Figure 2A), the CT scan of neck showed disappearance of tracheal compression and residual tumor in the left lobe of thyroid (Figure 2B). Then radiotherapy with a dose of 70Gy/35f to the tumor and 50Gy/25f to the related lymphatic drainage area was carried out. The neck CT scan of 3 month after radiotherapy demonstrated a stable

References

1. Cho JK, Woo SH, Park J, Kim MJ, Jeong HS (2014) Primary squamous cell carcinomas in the thyroid gland: an individual participant data meta-analysis. *Cancer Med* 3: 1396-1403.
2. Syed MI, Stewart M, Syed S, Dahill S, Adams C, et al. (2011) Squamous cell carcinoma of the thyroid gland: primary or secondary disease? *J Laryngol Otol* 125: 3-9.
3. Lam KY, Lo CY, Liu MC (2001) Primary squamous cell carcinoma of the thyroid gland: an entity with aggressive clinical behaviour and distinctive cytokeratin expression profiles. *Histopathology* 39: 279-286.
4. Chen KH, Chou YH, Cheng AL (2012) Primary squamous cell carcinoma of the thyroid with cardiac metastases and right ventricle outflow tract obstruction. *J Clin Oncol* 30: e260-263.
5. Lui JT, Khalil MN, Chandarana SP (2014) Primary squamous cell of the thyroid-an abbreviated clinical presentation. *J Otolaryngol Head Neck Surg* 43: 17.
6. Yucel H, Schaper NC, van Beek M, Bravenboer B (2010) Primary squamous cell carcinoma of the thyroid years after radioactive iodine treatment. *Neth J Med* 68: 224-226.
7. Jang JY, Kwon KW, Kim SW, Youn I (2014) Primary squamous cell carcinoma of thyroid gland with local recurrence: ultrasonographic and computed tomographic findings. *Ultrasonography* 33: 143-148.
8. Ko YS, Hwang TS, Han HS, Lim SD, Kim WS, et al. (2012) Primary pure squamous cell carcinoma of the thyroid: report and histogenic consideration of a case involving a BRAF mutation. *Pathol Int* 62: 43-48.
9. Batchelor NK (2011) Primary squamous cell carcinoma of the thyroid: an unusual presentation. *J Bronchology Interv Pulmonol* 18: 168-170.
10. Tunio MA, Al Asiri M, Fagih M, Akasha R (2012) Primary squamous cell carcinoma of thyroid: a case report and review of literature. *Head Neck Oncol* 4: 8.
11. Shrestha M, Sridhara SK, Leo LJ, Coppit GL 3rd, Ehrhardt NM (2013) Primary squamous cell carcinoma of the thyroid gland: a case report and review. *Head Neck* 35: E299-303.
12. Ghoshal S, Bhattacharyya T, Sood A, Tyagi R (2013) Palliative radiation in primary squamous cell carcinoma of thyroid: a rare case report. *Indian J Palliat Care* 19: 192-194.