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Case Report

A postoperative spindle cell tumor in the thyroid bed resembling recurrent papillary thyroid carcinoma: a 4.5-year follow-up case report

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Abstract: Postoperative spindle cell tumor involving the head and neck is relatively rare. Therefore, a differential diagnosis between local recurrence after thyroidectomy for papillary thyroid carcinoma (PTC) and postoperative spindle cell tumor is hard to come by. A 56-year-old man presented with a neck mass that was difficult to distinguish from local recurrence on neck ultrasonography (US) following previous successful total thyroidectomy and postoperative I-131 radioactive iodine (RAI) remnant ablation for PTC 34 months prior. Repeated US-guided fine needle aspiration and diagnostic I-131 RAI whole-body scan were inconclusive. Thyrotropin (TSH)-stimulated thyroglobulin (Tg) and F-18-fluorodeoxyglucose positron emission tomography-computed tomography results could not rule out the recurrence of PTC. The patient successfully underwent surgical removal of the neck mass. The final diagnosis was a postoperative spindle cell tumor in the thyroid bed. The patient showed no evidence of any recurrence or metastasis on follow-up monitoring with serum Tg and neck US for 4.5 years after resection of the mass. Our report might give further guidance in avoiding erroneous and missed diagnosis for the postoperative spindle cell tumor, which is different from recurrent papillary thyroid carcinoma, after total thyroidectomy and I-131 RAI.

Keywords: Postoperative spindle cell tumor, papillary thyroid carcinoma, case report

Introduction

To minimize the risk of recurrence of papillary thyroid carcinoma (PTC), including papillary thyroid carcinoma, complete surgical resection, radioactive iodine (RAI) ablation, and thyrotropin (TSH) suppression play important roles in patients [1]. After their initial treatment, PTC patients receive long-term surveillance for possible recurrence or metastasis with measurement of serum thyroglobulin (Tg) levels, high-frequency neck ultrasound (US), and diagnostic whole-body RAI scans (WBS). US is known to be a very sensitive surveillance modality, and US-guided fine needle aspiration (FNA) is recommended for accurate diagnosis of US-detected suspicious lesions (especially those ≥8-10 mm in size) in the postoperative thyroid bed. According to a retrospective hospital cohort study, the possibility of "non-diagnostic" findings has been reported in 33% following US-guided FNA [2].

For patients with PTC who underwent total thyroidectomy and RAI ablation, elevated serum Tg levels and negative results on I-131 WBS make it difficult to detect local recurrence or metastasis. Among various imaging modalities, F-18-fluorodeoxyglucose positron emission tomography-computed tomography (F-18 FDG PET-CT) plays a crucial role in diagnosing the recurrence of PTC by providing information regarding metabolic activity, especially in Tg-positive and non-RAI-avid cases [3, 4]. If the size of the suspicious lesion under surveillance continues to increase, a comprehensive diagnostic approach is required to improve PTC patients' survival while avoiding overtreatment.

Here, we report a case of a postoperative spindle cell tumor that mimics recurrent disease on US, F-18 FDG PET-CT, and detectable TSH-stimulated Tg in a patient who had undergone total thyroidectomy and RAI ablation therapy.

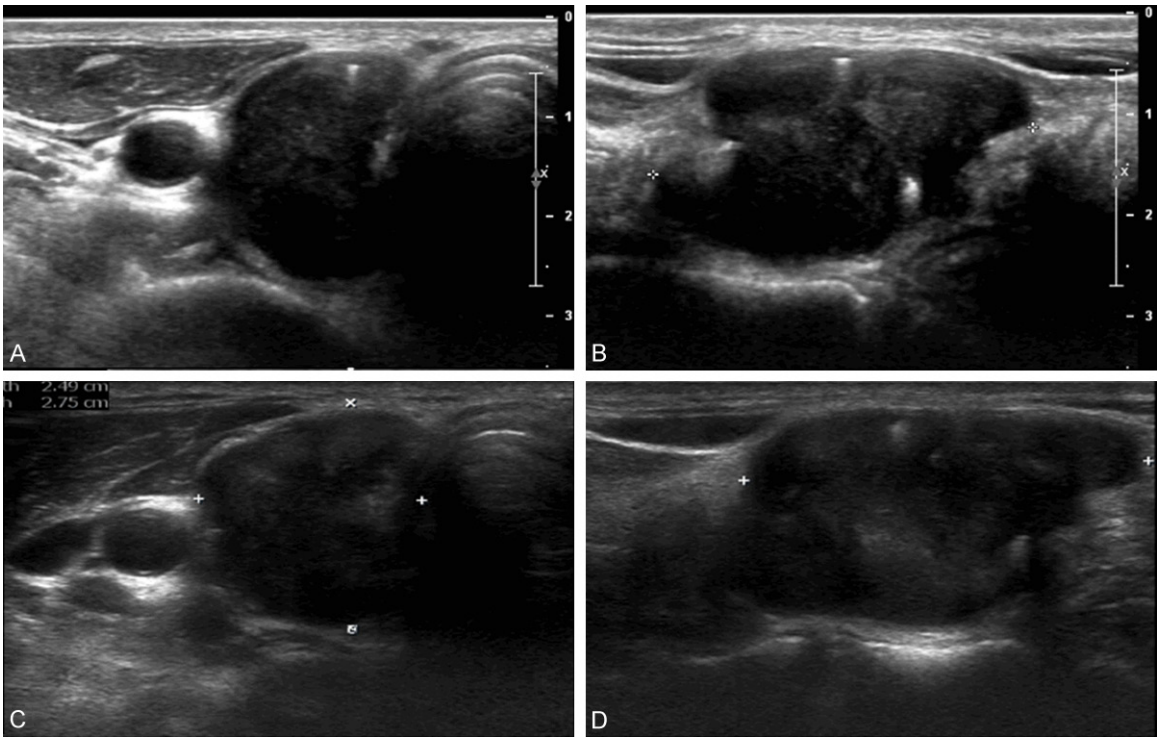

Figure 1. Neck ultrasonography. A and B. Neck ultrasonography (US) showed a hypoechoic mass (1.6×2.3×3.5 cm) with a lobulated margin and macrocalcifications in the postoperative right thyroid bed at 34 months postoperatively. C and D. After 1 month, the hypoechoic mass had increased in size (2.0×2.5×3.6 cm).

Case presentation

A 56-year-old man visited the endocrinology outpatient clinic, and a hypoechoic nodule in the right postoperative thyroid bed was found during regular check-up in April 2015. He had undergone total thyroidectomy and RAI remnant ablation with I-131 (80 mCi) for treatment of a papillary thyroid carcinoma. Initial AJCC 7th edition/TNM staging was III (T3aN1aM0). Twelve months after RAI remnant ablation, follow-up I-131 RAI WBS showed no evidence of thyroid remnants or metastatic lesions, and TSH-stimulated Tg was undetectable. During a follow-up with neck US, a hypoechoic solid mass measuring 1.6×2.3×3.5 cm in size with a lobulated margin and macrocalcifications in the postoperative right thyroid bed was observed. US-guided FNA was performed at the suspicious lesion (**Figure 1A** and **1B**). The smear of the FNA cytology showed cystic fluid only. After 1 month, the mass had increased slightly in size (2.0×2.5×3.6 cm), and a non-diagnostic result was obtained following repeated US-guided FNA (**Figure 1C** and **1D**). The cytological diagnoses were non-diagnostic or unsatisfactory (Bethesda system for reporting thyroid cytopathology category I). Non-stimulated serum Tg was undetectable during the follow-up period. To assess potential disease recurrence, TSH-stimulated serum Tg was checked. TSH-stimulated Tg was 2.15 ng/mL in the absence of anti-Tg antibodies. Due to the non-RAI-avid status, F-18 FDG PET-CT was performed to assess for its neoplastic focus. The maximum standardized uptake value (SUVmax) on F-18 FDG PET-CT was 2.4 at the mass, and a hypermetabolic lymph node was observed at the right cervical level IV (**Figure 2**). The neck CT revealed a focal decreased density at the right tracheal cartilage, suggesting tumor invasion (**Figure 3**). Given these imaging findings and the increased level of TSH-stimulated Tg, local tumor recurrence could not be ruled out; therefore, the patient underwent neck mass excision after providing informed consent. Adhesions on adjacent structures including the trachea and esophagus were observed. Grossly, the neck mass measured 4.0×3.0×2.0 cm in size, with a firm and gray-whitish surface. Microscopically, the mass was composed of spindle-

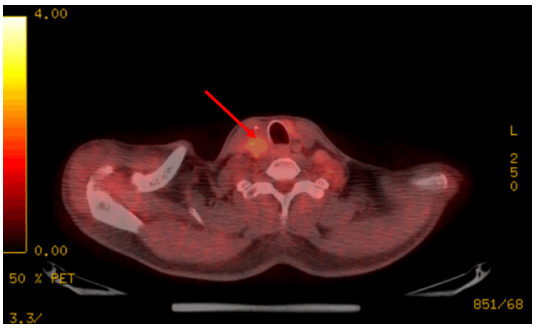

Figure 2. Whole body F-18 fluorodeoxyglucose positron emission tomography-computed tomography. The maximum standardized uptake value (SUVmax) was 2.4 at the mass and a hypermetabolic lymph node was observed at the right cervical level IV (as indicated by the red arrow).

shaped cells with eosinophilic cytoplasm, delicate blood vessels and focal myxoid change (**Figure 4**). The spindle cells were positive for vimentin, whereas they were negative for pan-cytokeratin (AE1/AE3), CD34, S-100 protein, α-smooth muscle actin (SMA), and desmin in immunohistochemistry (IHC) stains. The final pathological diagnosis was a postoperative spindle cell tumor, suggestive of myofibroma. The patient was monitored using non-stimulated Tg and neck US every 3 to 6 months. The evidence of tumor recurrence or metastasis was not found. At present, 4.5 years after excision of the postoperative spindle cell tumor, he continues to receive TSH suppression therapy without any complications.

Ethics statement

Informed written consent for publication of clinical data was obtained from the patient. The study was approved and monitored by the ethics committee of Chungbuk National University Hospital (IRB No. CBNUH 2020-02-016).

Discussion

Considering that most recurrent PTCs are detected in the neck, long-term follow-up of postoperative PTCs is mainly based on neck US, serum Tg after thyroidectomy, and I-131 RAI [1]. Although neck US is difficult to distinguish between recurrent and benign nodules at thyroid bed, the following findings should be considered as malignant: LN ≥8 mm in the shortest diameter, a heterogeneous echogenic, accompanied by calcification, a cystic appearance, a round shape, or the loss of the hyperechoic fatty hilum. Ultrasonographically suspicious lymph nodes 8-10 mm or more in the shortest diameter are strongly recommended to be US-guided FNA for cytology with washed Tg measurement [1]. Some recurrent PTCs are not easily detected, for example, the so-called "Tg-positive scan-negative" lesions are known to be difficult to detect. In 1981, Ashcraft and Van Herle described Tg-positive scan-negative disease in a group of PTC patients who were I-131 WBS-negative, with detectable serum Tg after thyroidectomy and I-131 RAI [5]. Detectable Tg levels were generally defined in previous studies as follows: TSH-stimulated Tg >5.0 to 10.0 ng/mL or TSH-nonstimulated Tg >1.0 to 2.0 ng/mL [5, 6]. A previous study reported that persistent lymph node lesions were detected in patients with TSH-stimulated Tg >1.0 ng/mL and negative for I-131 WBS [7]. Regarding a diagnostic approach for Tg-positive scan-negative patients, an early alternative imaging evaluation that provides clinicians with additional diagnostic clues, such as the F-18 FDG PET-CT scan, is needed.

Postoperative spindle cell nodules (PSCNs) are known as reactive, localized lesions that occur several weeks to months after a traumatic procedure or operation [8]. PSCNs have similar pathological characteristics to inflammatory myofibroblastic tumors and are reported to occur mainly in the genitourinary tract [9]. PSCNs are characterized by proliferative spindle-shaped cells with eosinophilic cytoplasm and large nuclei without hyperchromatism. Usually, PSCNs are clinically benign tumors that exhibit infiltrations of inflammatory cells on ulcerative surfaces, similar to sarcoma, but without atypical mitosis, necrosis or significant polymorphism. However, PSCNs of the thyroid are rare so that they may be overlooked [10]. A previous study reported 10 cases of PSCNs (1-7 mm in size) in patients who had undergone FNA that could be misdiagnosed as anaplastic carcinoma, medullary carcinoma, or sarcoma [11]. In our case, the IHC staining for vimentin was positive, but there were negative results for pan-cytokeratin (AE1/AE3), CD34, S-100 protein, SMA, and desmin immunostaining. Regarding the differential diagnosis of PSCNs, IHC analyses of vimentin, SMA, cytokeratin, and desmin are

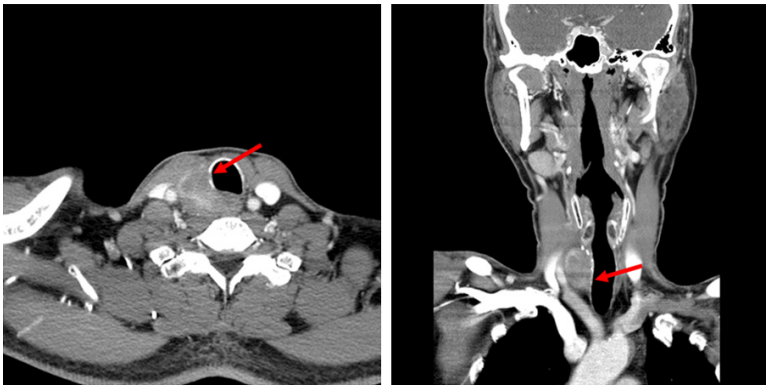

Figure 3. Computed tomography of the neck. A focal decreased dense mass at the right tracheal cartilage (3.1×2.1×2.4 cm) was observed, suggesting tumor invasion (as indicated by the red arrows).

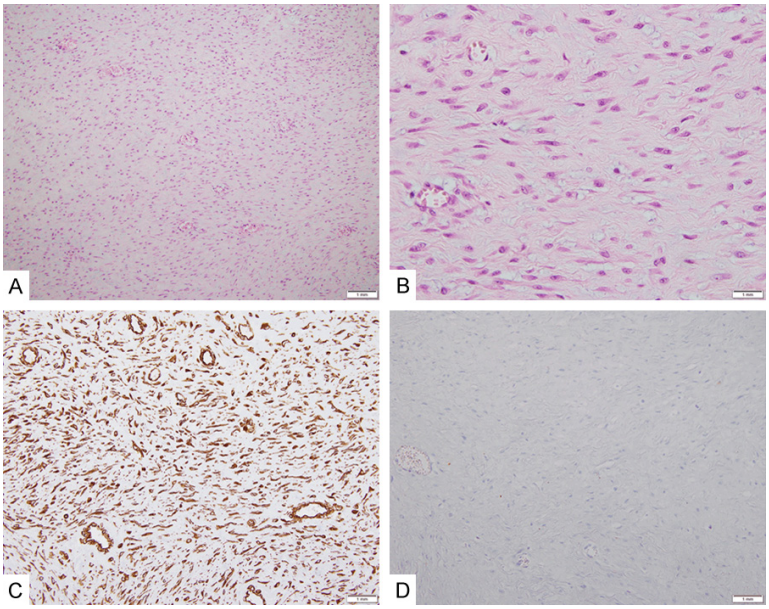

Figure 4. Histopathology and immunohistochemistry of the postoperative spindle cell tumor. (A) and (B). Proliferation of bland-looking spindle cell with delicate blood vessels and focal myxoid change were observed (hematoxylin-eosin stain; original magnification, ×100, ×400, respectively). (C). Spindle cells are positive for vimentin but negative for pan-cytokeratin (AE1/AE3) (D) in immunohistochemistry.

recommended useful studies [12]. Previous study reported that the IHC stains for desmin, and cytokeratin are inconclusive because of their lower positivity [13]. In some studies, p53 IHC stain is used as an additional differential diagnosis between PSCNs and malignant tumors; negative results were reported as PSCNs and positive results suggested more malignant potential [14, 15]. Our case exhibited positive IHC staining only for vimentin, which is common in typical PSCNs, and a clinically benign lesion without evidence of metastasis or recurrence over the 4.5 years was present.

Conclusion

We reported a case of postoperative spindle cell tumor in the thyroid bed that mimicked recurrent thyroid cancer on neck US, TSH-stimulated Tg, and F-18 FDG PET-CT. This case highlights the importance of considering the differential diagnosis of postoperative spindle cell tumors in the thyroid bed in patients with papillary thyroid carcinoma who have undergone thyroidectomy.

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Disclosure of conflict of interest

None.

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