

Metastatic Spinal Cord Compression as Initial Presentation of Occult Follicular Thyroid Carcinoma

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Spinal cord compression is a rare complication of differentiated thyroid carcinoma (DTC), which may present late in the course of the disease. Spinal cord compression as the initial presentation of DTC is an extremely rare condition. We present a case of follicular thyroid carcinoma in a patient who presented for an evaluation of spinal cord compression. A thorough search of the literature revealed only 15 similar cases. We summarize the clinical characteristics, therapeutic regimens, outcomes, and prognoses of these cases. Additional management issues are also discussed, with a thorough literature review.

Key words: spinal cord compression, follicular thyroid carcinoma, differentiated thyroid carcinoma

INTRODUCTION

Thyroid cancer constitutes 1%-2% of all malignant neoplasms¹ and follicular thyroid carcinoma constitutes 15%-30% of all malignant thyroid neoplasms². Bone is the second most common site of metastasis resulting from thyroid cancer, after the lung³. In contrast, follicular thyroid carcinoma is the most common histological origin of bone metastasis among differentiated thyroid carcinomas (DTCs), with an incidence ranging from 7% to 28%^{4,5}. Metastasis of a malignancy to the bone, specifically to the vertebral column, may present as bone pain, pathological fracture, or cord compression, and is frequently a surgical issue⁶. However, spinal cord compression, as a complication of thyroid carcinoma, is uncommon and occurs mainly during the late stages of the disease, resulting from a local extension or hematogenous spread of the primary tumor.

We present here a rare case of a patient with occult follicular thyroid carcinoma that manifested initially as spinal cord compression. We also review the literature on

Received: June 11, 2007; Revised: October 22, 2007; Accepted: October 31, 2007

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other similar cases and the principles of management.

CASE REPORT

A 55-year-old woman presented at our orthopedic clinic with a two-month history of sharp pain in the lumbar region, radiating to both lower extremities. The pain was exacerbated by standing or walking and improved with sitting. She also experienced numbness on the lateral aspects of both calves and feet. An initial trial of nonsteroidal anti-inflammatory drugs resulted in mild improvement in her symptoms until one week before admission, when she was unable to stand, and developed bowel and bladder incontinence. The patient had undergone a subtotal hemithyroidectomy at another hospital 10 years earlier, with histologically reported benign follicular adenoma. Her medical history was otherwise unremarkable.

On physical examination, there was a marked loss of muscle function, with grades 1 and 4 strength in the left and right lower extremity muscles, respectively. A magnetic resonance imaging (MRI) scan showed bony destruction of the fifth lumbar vertebra and sacrum by a $10 \times 9 \times 12$ cm³ mass lesion, with total obliteration of the adjacent spinal canal (Fig. 1). An excisional biopsy of the lesion was performed and a pathological examination revealed a metastatic follicular carcinoma originating from the thyroid gland (Fig. 2). The patient then underwent a total thyroidectomy, but no tumor was found in the extirpated tissue. Unfortunately, no tissue samples from the previous



Fig. 1. T1-weighted sagittal magnetic resonance imaging of the vertebral column. A 10 x 9 x 12 cm mass lesion (arrows) caused bony destruction of the fifth lumbar vertebra and sacrum with total obliteration of the spinal canal at this level.

hemithyroidectomy were available for review. She was subsequently treated with an ablation dose of 150 mCi ¹³¹I.

One month later, she returned with full paraplegia, lower extremity edema, and worsening pain extending from the lower back to both lower extremities. Repeated MRI scans revealed no reduction in the tumor size, and the fourth lumbar vertebra was involved. Moreover, the tumor had induced moderate bilateral hydroureteronephropathies, followed by an episode of acute pyelonephritis. She underwent percutaneous nephrostomies and received broadspectrum antibiotics. External-beam irradiation was offered, but she declined. She was therefore admitted for palliative care. She died one month later from urosepsis as a consequence of progressive metastatic disease.

DISCUSSION

Distant bony metastasis, as a complication of DTC, is not rare, occurring mainly in the late stages of the disease. However, spinal cord compression as an initial manifestation of newly diagnosed thyroid carcinoma is a rare event. Shaha et al.⁷ reported that only 44 of 1038 (4%) patients with thyroid cancer presented initially with a distant metastasis. Of all thyroid cancer subtypes, follicular carcinoma is the most likely to present with distant metastasis⁷ or to do so as a late event in a long-standing disease⁸. Fornasier

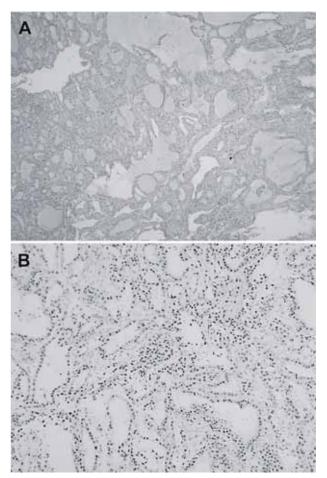


Fig. 2. Metastatic follicular carcinoma originating from the thyroid gland in the excised spinal tumor specimen was found, which composed of a varying-sized follicular arrangement of tumor cells with colloid contents (A, H&E, original magnification x100) and positive for thyroid transformation factor 1 by immunohistopathologic staining (B, TTF-1, original magnification ×200).

and Horne⁹ reviewed 374 autopsy specimens from patients with malignancies, 140 of whom had metastatic spread to the vertebral bodies. Only one thyroid carcinoma that had metastasized to a vertebral body was identified. Barron et al.¹⁰ reviewed 127 autopsies of patients with spinal cord compression resulting from metastatic neoplasms, and only three of the tumors were of thyroid origin.

Available data indicate that only a few sporadic cases of spinal cord compression presenting as the initial manifestation of occult thyroid carcinoma have been reported (Table 1)¹¹⁻²⁰. The most common type among these cases was follicular carcinoma, which is consistent with the findings of Shaha et al.⁷. Although some patients underwent only surgical treatment and/or external irradiation for

Table 1 Metastatic Thyroid Carcinoma Presenting Initially With Spinal Cord Compression

No.	Gender/Age	Histology	Spread	Location	Therapy	Survival*	Outcome
1	M/76	Papillary	Direct extension	C5-C7	RT	1 wk	DOD
2	M/57	FTC	Metastasis	T5-T6	Laminectomy, excision, thyroidectomy, 100 mCi I-131	1 yr	NED
3	M/56	FTC	Metastasis	L2+C6	Thyroidectomy, 148 mCi I-131	17 yr	Die of leukemia
4	M/62	Papillary-follicular	Metastasis	T2-T3	RT, I-131 ablatio (doseunknown)	8 yr	NED
5	M/58	Papillary-follicular	Metastasis	T10	Laminectomy, excision, thyroidectomy, 196 mCi I-131	27 mo+	NED
6	F/65	FTC	Metastasis	C2	Resection, thyroidectomy, I-131 ablation	28 mo	NED
7	F/43	FTC	Metastasis	T1	RT, thyroidectomy	10.5 mo	DOD
8	M/50	FTC	Metastasis	C6	Laminectomy, thyroidectomy, RT, I-131 ablation	Unknown	NED
9	M/72	Insular carcinoma of thyroid	Metastasis	C6-C7	Laminectomy, thyroidectomy, RT, I-131 ablation	14 mo+	NED
10	M/60	FTC	Metastasis	C7+T7+L2	RT, laminectomy, thyroidectomy, embolization, 150 mCi I-131	6 yr+	NED
11	M/69	Insular type FTC	Metastasis	L3-L4	Thyroidectomy, embolization, RT, laminectomy	2yr+	NED
12	M/71	Papillary (follicular variant)	Metastasis	T2	RT, thyroidectomy, laminectomy	6 yr+	NED
13	M/61	FTC	Metastasis	Cervical, thoracic and sacral regions	Succumbed before therapy of FTC	-	Die of pneumonia
14	F/53	FTC	Metastasis	L5-S1	RT, thyroidectomy, laminectomy, 200 mCi I-131	6 yr+	NED
15	F/61	FTC, papilla (follicular variant)	Metastasis	T1-T2	Resection, thyroidectomy, 100 mCi I-131	Unknown	NED
16	F/55	FTC	Metastasis	L5, sacrum	Thyroidectomy, 150mCi I-131	2 mo	DOD Present case

^{*}Since end of treatment; FTC, follicular thyroid carcinoma; RT, radiation therapy; DOD, die of disease; NED, no evidence of disease

the relief of symptoms, an appropriate and intensive treatment of both the metastatic and primary thyroid tumors is required to achieve long-term survival and a good quality of life for the patients.

To prevent further neurological deficits, it is usually advisable to initially and promptly stabilize the spine, especially in the context of potential long-term survival. Traditionally, corticosteroids, local radiation treatment, and surgery to the vertebrae were all thought to be important for most patients with spinal cord compression. Stojadinovic et al. 10 recommended surgery as the preferred method for resectable, locoregional recurrence, followed by radioactive iodine (RAI) therapy for iodide-concentrating thyroid cancer, or external-beam radiation for tumors that lack RAI avidity. Those investigators also found that complete palliative debulking of the localized metastatic lesions of DTC may be associated with an improvement in the patient's survival and quality of life. However, solitary distant metastases of DTC are very rarely amenable to

complete resection, and thus some local procedures to delay tumor progression and for symptom palliation are used, such as embolization, radiofrequency, or cement injection and treatment with biphosphonates²¹. In two cases listed in Table 1 (cases 10 and 11), the most significant proof of the effectiveness of embolization was the immediate relief of the neurological symptoms 18. Furthermore, Smit et al.¹⁸ recommended that selective embolization is an attractive strategy for the treatment of symptomatic vertebral metastasis from a thyroid carcinoma because it is less invasive than surgery and offers faster relief of symptoms than does RAI therapy. However, the management of DTC patients with distant metastases is still controversial, especially in terms of the management of bone metastases²²⁻²⁵. Recently, Chow et al.^{22,23} reported that RAI therapy is the single independent prognostic factor for survival in patients with distant metastases. The five-year survival rates for patients with and without RAI therapy were 49% and 15%, respectively²³. Moreover,

both the locoregional recurrence rate and the mortality rate were reduced to about 25% in patients treated with RAI²². A recent review of the literature indicated that although external radiotherapy in association with RAI therapy has an effect on cancer recurrence, pain relief, and the recalcification of osteolytic lesions, external radiotherapy per se cannot improve the survival rate²⁶. Instead, complete removal of any tumoral bone tissue in patients less than 45 years old and a cumulative dose of RAI therapy appeared to improved survival in patients with bone metastases originating from DTC²⁴.

The prognosis of occult thyroid carcinoma with distant metastasis also remains a source of contention. Proye et al.²⁵ demonstrated that DTC is usually less life threatening, and that early diagnosis and appropriate treatment for distant metastases can significantly prolong the life span and improve life quality. Shaha et al. also reported that total thyroidectomy followed by RAI therapy and thyroxinesuppressive treatment extended long-term survival (10-15 years) in 44% of patients with metastatic follicular thyroid carcinoma. However, Pittas et al.²⁷ reported that the 10year survival of patients diagnosed with bone metastasis was only 13%. It is interesting to note that three patients in Table 1, including the patient presented herein, had survival times of less than two months. We believe that, with the exception of older age at presentation (e.g., case 1 was > 70 years of age), which has a major impact on the prognosis, patient refusal of further treatment (the patient described herein refused radiotherapy and further RAI therapy) and the inevitable significant comorbidities (case 13 had pneumonia; the case presented here had obstructive nephropathy followed by urosepsis) probably have a major adverse effect on survival.

In conclusion, although it is rare, thyroid carcinoma should be considered in the differential diagnosis of every patient with new-onset spinal cord compression, an emergency situation calling for prompt diagnosis and treatment. We believe that the management of this medical emergency should be a multidisciplinary effort. Prompt management of the primary carcinoma and the metastatic lesion, ongoing maintenance of thyroid suppression, and consideration of the patient's age, response to therapies, and comorbidities, may extend long-term survival and allow a favorable prognosis.

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