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SURGICAL NEUROLOGY INTERNATIONAL

SNI: Spine, a supplement to *Surgical Neurology International*

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Intramedullary spinal cord metastasis arising from papillary thyroid carcinoma: A case report and review of literature

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Received: 27 January 16 Accepted: 01 April 16 Published: 17 May 16

Abstract

Background: Intramedullary spinal cord metastases (IMSCM) are typically drop lesions from intracranial metastases and are a rare manifestation of systemic malignancy (8.5% of central nervous system metastases). They arise from primaries such as the lungs, breast, kidney, melanoma, or lymphoma. On the other hand, they arise very rarely from papillary thyroid carcinoma (PTC), even though it is the most common type of primary thyroid malignancy.

Case Description: A 54-year-old male presented with pain in the lower back along with tingling, numbness, and weakness in the bilateral lower limbs. This was associated with urine incontinence for 1½ months. In the previous month, he developed a left-sided solitary thyroid nodule. Fine needle aspiration cytology and ultrasonography were suggestive of metastasis. Furthermore, the thoracolumbar magnetic resonance imaging showed T1-hypo and T2-hyper-intense D11-D12 level intramedullary lesion, with intense enhancement, which was consistent with an intramedullary lesion involving the conus. At surgery, a firm, brownish yellow, friable, vascular tumour was removed *en toto*. Upon discharge, the patient was neurologically intact except for residual bladder incontinence.

Conclusion: In an extensive literature review (pubmed), IMSCM metastasis from PTC primary is confirmed as a rarity and this may be the fourth documented case. Moreover, this may be the first report of a case of PTC metastatic neurological deterioration "even before the treatment of the primary was undertaken." Early diagnosis and microsurgical resection can result in improvement of neurological deficits and in the quality of life of patients with IMSCM.

Key Words: Intramedullary spinal cord metastasis, laminectomy, papillary thyroid carcinoma, spinal metastasis, thyroid cancer

Access this article online

Website: www.surgicalneurologyint.com

DOI: 10.4103/2152-7806.182544

Quick Response Code:

How to cite this article: Tripathy SR, Deo RC, Mishra S, Dhir MK, Nath PC, Satapathy MC. Intramedullary spinal cord metastasis arising from papillary thyroid carcinoma:A case report and review of literature. Surg Neurol Int 2016;7:S375-9.
http://surgicalneurologyint.com/Intramedullary-spinal-cord-metastasis-arising-from-papillary-thyroid-carcinoma:-A-case-report-and-review-of-literature/

INTRODUCTION

Intramedullary spinal cord metastases (IMSCM) represent 8.5% of central nervous system (CNS) metastases. IMSCM affects 0.1–0.4% of all carcinoma patients and comprise 1–3% of all intramedullary spinal cord neoplasms.[6-8] Most of these tumours are drop lesions from intracranial metastases and are attributed to primaries such as lungs, small cell carcinoma being the most common, with the rest originating from primaries in breasts, kidneys, melanoma, or lymphoma.[10] IMSCM from thyroid cancer is extremely rare and only a few cases have been reported (e.g., most found during autopsy). In this report, the authors present a patient with an IMSCM arising from papillary thyroid carcinoma (PTC). Although PTC is the most common histological type of thyroid malignancy with an increasing incidence in many countries, its involvement as an etiology for IMSCM is rare. In an extensive literature review (pubmed), it was confirmed that this is probably the fourth case report of IMSCM due to thyroid primary and the first reported case of IMSCM-related neurological deterioration, even before the primary pathology is dealt with.

CASE REPORT

A 54-year-old male presented with bilateral low back pain, tingling, numbness, and weakness in both the lower limbs, and incontinence of the bowel and bladder of 2-months duration [Figure 1]. In the month prior to the presentation, he developed an anterior neck mass. Fine needle aspiration cytology (FNAC) suggested a PTC and the ultrasound of the neck showed an enlarged thyroid gland (the largest cyst in the left lobe, with calcification and increased vascularity), with multiple enlarged, necrotic cervical nodes, which was suggestive of cervical metastases.

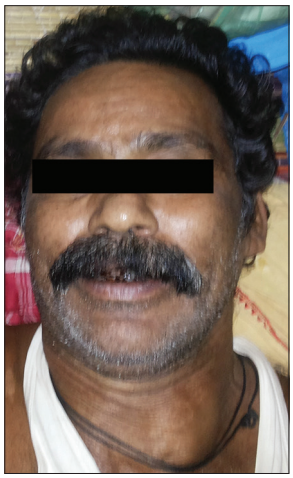

Figure 1: The patient had a left anterior neck mass, which showed on fine needle aspiration cytology as papillary thyroid carcinoma. Ultrasonography of the neck did not show any other neck nodes

On examination, he had a solitary left-sided thyroid nodule (2 × 2 cm). Motor strength in the lower extremities was 4-/5 on the left and 4/5 on the right (Medical Research Council scale). There was a 20% diminution of all sensation below L1 dermatome and 40% below L3. Perianal sensation was diminished. Plantar reflex was extensor on the left and flexor on the right side.

MR studies

At the D11-12 level, magnetic resonance imaging (MRI) showed an expanded cord with an intramedullary lesion (13 × 11 × 8 mm); the T1 image was hypointense and the T2 study showed it was hyperintense with intense enhancement [Figure 2]. The working radiological impression was that of a hemangioblastoma involving the conus medullary segment.

Surgical intervention

A midline vertical incision was made along the D10-L1 spinous process. D11, D12 laminectomy was performed revealing a tense bluish, intradural mass within the dura in the conus region [Figure 3]. On longitudinal durotomy, the tumor was found to be brownish yellow, soft, easily aspirated, vascular, and friable. The tumor was removed *en toto*.

Postoperative neurological status

On postoperative day 3, the patient could walk with minimal support, and could resume his daily activities within a week. However, his bladder incontinence persisted, requiring a perurethral catheter *in situ* during discharge.

Pathology

H and E (hematoxylin and eosin) staining showed neoplastic tissue composed of sheaths of cells with ovaloid

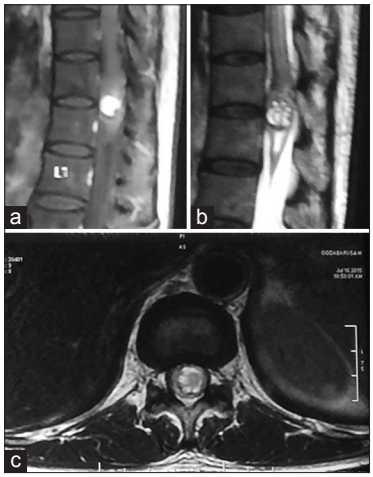

Figure 2: The MRI scan of the thoracic spinal cord: Sagittal (a) T1-weighted contrast image, sagittal (b) T2-weighted image, axial (c) T1-weighted contrast image and axial: Showing a solitary mass in the intramedullary spinal cord at D11-12 level. The lesion showed a heterogeneous enhancement following intravenous gadolinium contrast

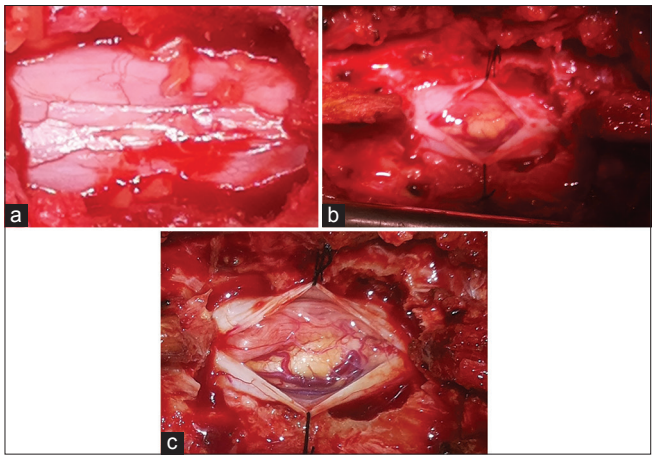

Figure 3: The operative picture as seen under operating microscope (a) before durotomy and (b) after durotomy; (c) the brownish yellow, soft to firm, suckable, vascular, friable tumour

nuclei and without a cytoplasmic border; some had a ground glass appearance and nuclear grooves. In addition, there were many unevenly distributed psammoma bodies between the cells. Immunohistochemically, tumour cells were strongly positive for cytokeratin (CK), CK7, thyroid transcription factor 1 and negative for Novel Aspartic Proteinase of the PepSIN Family type A (NAPSIN-A) and CK20. Pathology confirmed metastatic lesion from PTC.

DISCUSSION

IMSCM are uncommon neoplastic lesions and account for 3–5% cases of myelopathy in patients affected by carcinoma.[7] As many as 2% of patients in the end stage of disseminated cancer are found to have spinal cord metastasis (SCM).

PTC is the most common type of thyroid tumour comprising between 30% and 70% of all thyroid carcinoma cases.[4] It usually remains intrathyroidal and tends to spread via local extension or metastasis to regional lymph nodes. Distant metastases are very uncommon and mainly occur in lungs and bones and almost never to the CNS.

Pathways for spread

Five routes have been hypothesised for the dissemination of metastatic intradural tumour originating from outside of the CNS: Haematogenous, via the arterial system, via the perineural lymphatics, through the venous plexus, spreading via subarachnoid space, and seeding from the involved osseous structure to the cerebrospinal fluid through the duramater.[10]

Clinical findings

Weakness and pain present early in comparison to sensory loss, with sphincter disturbance originating last.[13] It is almost impossible to make a positive diagnosis because the symptoms of hematomyelia are similar. Bleeding from thyroid carcinoma is uncommon. Anaplastic thyroid carcinoma (ATC) comprises only 1–3% of all thyroid neoplasms.[1,4] Bleeding from an ATC is a rare but potentially life-threatening complication.

The occurrence of metastatic lesions with hemorrhage arising from a PTC are extremely rare.[5] Spontaneous hemorrhage was presumably responsible for the acute progress of myelopathy in our patient.

Spinal cord apoplexy

Spinal cord apoplexy is a hemorrhage occurring directly into the spinal cord. It may occur due to traumatism, changes in the coats of the blood vessels, or excessive blood pressure. When its occurrence is recent, it is red or dark in colour, which changes to brown or yellow with age; the tumour in our patient had yellowish-brown discoloration. The clot may later become encapsuled by a fibrous deposit.[13] If the hemorrhage is small in quantity, absorption may speedily occur with complete recovery.[9] Prognosis depends upon the extent and location of the clot.

One report describes the case of a thoracic spinal cord lesion proven to be a metastasis from a thyroid primary (PTC).[11] The occurrence of metastatic lesions with hemorrhage arising from a PTC is extremely rare.[5] A few cases of lower gastrointestinal metastatic hemorrhage have been reported, which were associated with familial adenomatous polyposis.[15] Honma *et al.*[16] reported a case of intramedullary metastasis of presumed poorly differentiated thyroid carcinoma associated with a haemorrhage caudal to the tumour, which was identical to the configuration demonstrated in our patient.

Investigations

Gadolinium-enhanced MRI is the imaging modality of choice for spinal tumours, including spinal metastases.[7] Visualisation of the entire vertebral axis in multiple orthogonal planes is important given the high incidence of multiple vertebral levels of tumor deposits. MRI alters therapeutic decisions in a significant number of patients, especially with regard to the addition or modification of radiotherapy.[8]

Treatment

Providing patients with successful palliation and improving their quality of life demands multidisciplinary strategic treatment planning,[2,12,14] consisting of immediate steroid bolus, chemoradiotherapy, cytoreductive surgery, and radioactive iodine treatment.[17]

a) High dose dexamethasone allows for limited and transient neurological improvement
b) In combination with chemotherapy, radiation correlates with an increased duration of survival. Therapeutic irradiation and surgical decompression are the principal and complementary treatment options, respectively

Table 1: Reported similar scenarios - [IMSCM from PTC]

S. No (age/sex)	Neurological deficits			T/t History		Lab	MRI	FDG PET, or Tc99m	Surgery			Other Mx
	Motor	Sensory	Autonomic/ reflex	Δ	Intervention				Approach	Gross	H/P	Radioiodine
1. (31/male)[16]	Acute onset of paraplegia	Hypoesthesia below the umbilicus	Negative Anal tone, bilateral Babinski +ve	PTC	Thyroidectomy- 2 year back	Low TSH	33 mm mass at the D7-8, T1 iso, T2 Hyper intense, enhancing homogeneously	-	Laminectomy D6, 7, 8.	Dark-gray, yellowish	Metastatic papillary carcinoma	-
2. (71/female)[2]	C) Progressively increasing lower extremity weakness	A) Painful left sacroiliac area B) Recurrence in cervical lymph nodes C) Mid-thoracic burning pain radiating toward anterior chest wall	-	PTC- 6 years	A) Total thyroidectomy and hormone replacement therapy B) Surgery and 200 mCi iodine therapy	-	C) D4-D5 IDEM well defined, oval, T1 iso, T2 Hyper intense, marked homogenous enhancement	B) Increased uptake L5 and left sacroiliac joint C) No uptake in thoracic vertebrae	C) D3-D5 laminectomy	C) Solid greyish red mass intradural appeared compressing the cord	-	A) 100 mCi iodine ablative therapy B) Local external beam radiation therapy with 4000cGY
3.(44/female)[18]	Lower leg progressive weakness	A) 1997-bilateral lower leg pain		A) PTC- large mass in the right neck B) 2002- brain metastasis	total thyroidectomy			2×6 cm intramedullary mass, enhanced by gadolinium with FDG-uptake	Emergency laminectomy with subtotal tumor removal		Metastatic PTC	A) Repeated I131 therapies

c) Cytoreductive surgery is a major treatment option for lesions early in the course of neurological deficits; however, it has not been found to increase the duration of survival compared to other treatment modalities. In young patients, both PTC and follicular thyroid cancer (FTC) have a more than 97% cure rate if treated appropriately. Indications of surgery include failure of irradiation therapy, unknown diagnosis, pathological fracture dislocation, and rapidly progressing paraplegia[13]

d) Postoperatively, most, but not all, patients with thyroid carcinoma need radioactive iodine treatment, except medullary thyroid cancer (MTC) and some small PTC.[3]

Prognosis

The best predictor of outcome is the site of primary tumour type, with the best prognosis noted in patients with breast or kidney cancer. Life expectancy is often relatively short with median survival ranging from 4 to 15 months in various series.[8] Prognosis of IMSCM is compounded by the treatment modality and tumour histology with lung metastases correlating with the shortest survival.

Table 1 presents the prior reported cases of PTC primary giving rise to IMSCM.

CONCLUSION

Metastasis to spinal canal without the primary PTC manifesting itself is not common. In our extensive database search (Pubmed), this was confirmed to be the fourth such report of an IMSCM arising from a PTC. This may also stand out as the first report of a case of PTC metastatic neurological manifestation even before the treatment of the primary was undertaken. Early diagnosis and prompt microsurgical resection results in improvement of neurological deficits as well as the quality of life.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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