

Surgical Treatment for Cervical Lamina Metastasis from Parathyroid Carcinoma: A Case Report

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Parathyroid carcinoma (PC) is a rare malignancy, affecting merely 6 cases per 10 million individuals annually¹⁾. While it exhibits a propensity for wide dissemination, spinal metastases are relatively uncommon. In this report, we present the first case of spinal lamina metastasis from PC and elucidate its distinct clinical manifestations as well as the outcomes of surgical treatment.

A 53-year-old man experienced a recurrence of high calcium and parathyroid hormone (PTH) levels 28 months after *en bloc* resection for PC. His serum calcium level rose to 13.7 mg/dL, raising concerns regarding a possible recurrence (Fig. 1). He did not have subjective complaints of body pain or numbness. Neurological assessments were normal. However, positron emission tomography/computed tomography (PET/CT) scans showed abnormal uptake only in the 7th cervical lamina, indicating lytic bone lesions on CT. Furthermore, magnetic resonance imaging (MRI) revealed tumor lesions with isointensity on T1-weighted images, high intensity on T2-weighted images, and gadolinium enhancement on contrast-enhanced T1-weighted images (Fig. 2). Because medications including calcimimetics did not effectively control the high calcium levels (Fig. 1), it was decided to surgically remove the metastatic bone tumor.

The surgical procedure, which was performed with the patient in the prone position using a right-sided unilateral approach, commenced with tumor identification. The lamina and intervertebral joints around the tumor were excised. Although wide tumor resection was ideal, marginal resection was performed to protect the dura mater adjacent to the tu-

mor (Fig. 3).

Histopathological analysis revealed the solid growth of acidophilic round cells upon hematoxylin and eosin staining as well as positive PTH staining, confirming the diagnosis of bone metastasis originating from the PC. After stabilization of the calcium levels, the patient was discharged 12 days following surgery. Because the tumor in proximity to the dura was marginally resected, the patient underwent postoperative adjuvant radiation therapy with a total dose of 50 Gy 2 months following surgical resection. The MRI image taken 6 months postsurgery showed the absence of any residual tumor, and the patient exhibited stable calcium levels (Fig. 1); radiographic assessments indicated sustained alignment of the cervical spine without any evidence of recurrence at the 2-year follow-up.

We conducted an extensive literature search on spinal metastases of PC and found only seven reported cases: four cases in the lumbar²⁻⁵⁾, one in the thoracic⁶⁾, and two in the cervical spine^{7,8)} (Table 1). These reports showed that neurological manifestations or localized pain secondary to tumor infiltration were not common in cases of distant metastases from PC. Instead, six out of eight cases were identified by manifestations associated with hypercalcemia²⁻⁸⁾. Therefore, subsequent postoperative monitoring in PC emphasizes surveillance of hypercalcemia-related symptoms alongside regular hematological assessments. In cases of such hypercalcemia, recurrence or metastasis is suspected, and a systemic imaging search followed by therapeutic intervention should be conducted. However, we need to pay attention before the

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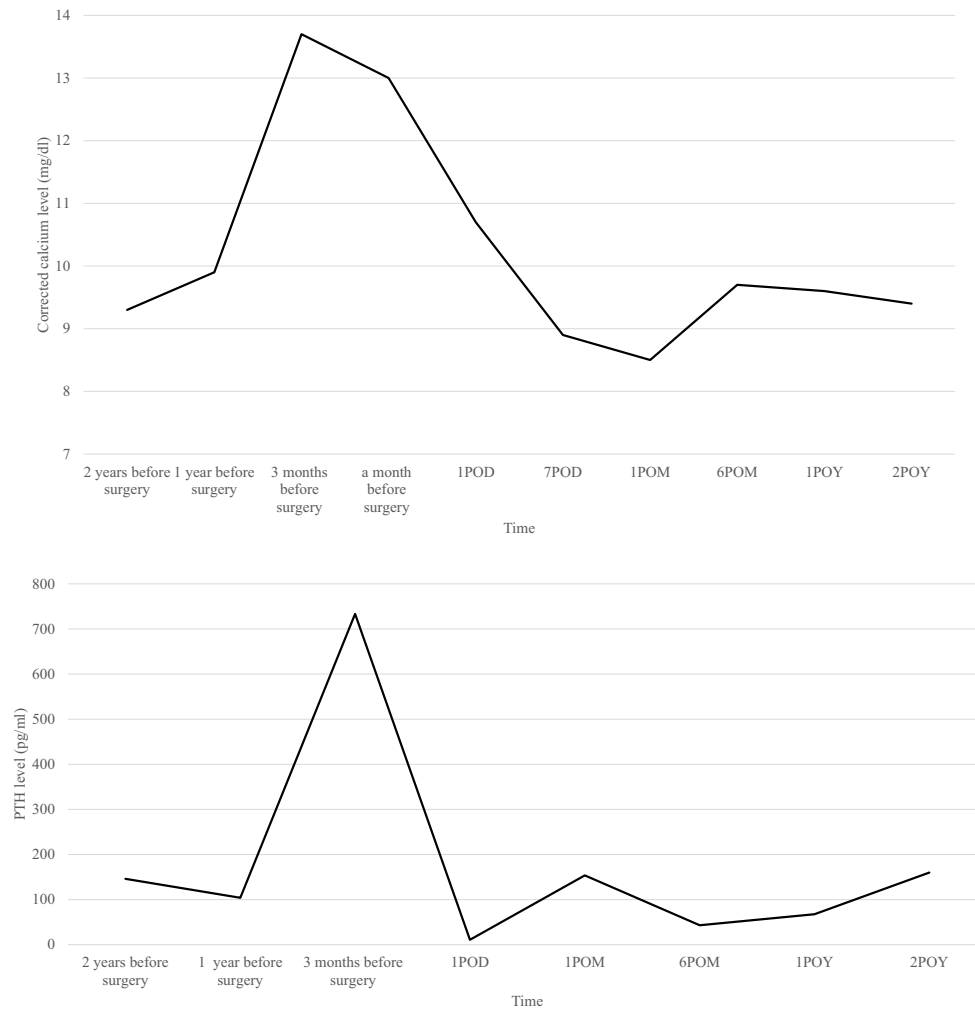


Figure 1. Corrected Calcium and PTH Levels Before and After Surgery.
 POD: Postoperative day, POM: Postoperative month, POY: Postoperative year
 3 months before surgery: Initial spike in serum calcium levels during follow-up.
 1 month before surgery: Last calcium level measured before surgery.

treatment is initiated as high PTH can cause benign osteolytic changes called Brown tumors, which have imaging findings similar to those of bone metastases⁹). Thus, we occasionally encountered cases of misdiagnosis, where Brown's tumor was mistakenly identified as bone metastases.

The treatment approach for PC heavily revolves around addressing parathyroid hormone-induced hypercalcemia, as it significantly influences the prognosis. In previous reported cases, except for a case where a residual tumor was observed, complete resection effectively controlled elevated PTH levels²⁻⁸). Thus, proactive surgical intervention for spinal metastases originating from PC remains pivotal even in asymptomatic cases. Regarding adjuvant therapy, the effectiveness of postoperative radiotherapy for PC remains uncertain, but it could be beneficial for carefully selected patient groups. However, due to the rarity of this condition, conducting larger studies to assess potential benefits may be difficult¹⁰).

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Interpretation of data: Masahiro Matsuda and Narihito Nagoshi.

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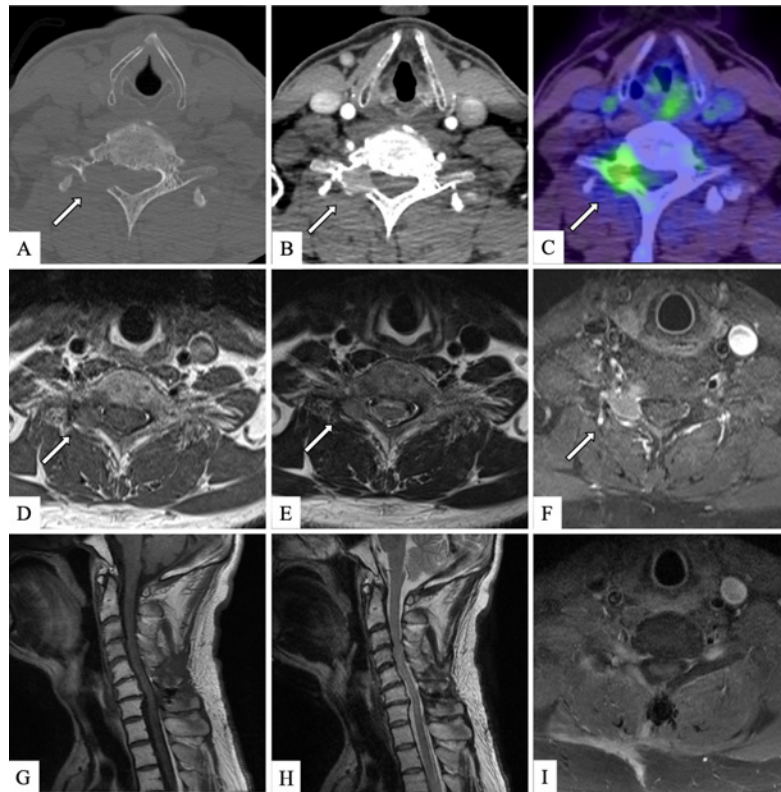


Figure 2.

Preoperative imaging includes bone-specific CT (A) and contrast-enhanced CT (B) in the axial view at the C7 vertebral level, showing a conspicuous mass shadow, indicative of bone tumor. PET/CT scan shows tracer accumulation localized within the right lamina of C7 (C). In addition, preoperative axial MRI at the C7 level consists of T1-weighted (D), T2-weighted (E), and contrast-enhanced T1-weighted sequences (F). T1 exhibits iso-signal, T2 displays high signal, and contrast-enhanced T1 reveals a mass originating from the lamina with partial invasion into the vertebral body. Postoperative MRI at the C7 level, taken 6 months after surgery, comprises sagittal T1-weighted (G), T2-weighted (H), and axial contrast-enhanced T1-weighted sequences (I).

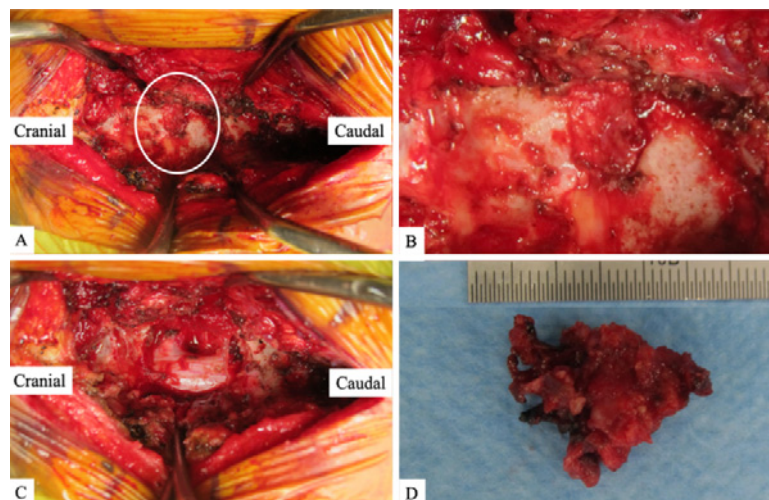


Figure 3.

Intraoperative images depict tumor lesion postdeployment using a right unilateral approach (A), visibly recognizable under direct observation. Detailed close-up view of the tumor site (B) and postresection images (C) are documented. Postresection image of the tumor measuring 20×16 mm (D).

Table 1. Spinal Metastasis of Parathyroid Carcinoma Previously Reported in the Literature.

Author	Year	Occurrence	Symptoms	Treatment	Outcome	Follow-up
Koyano H ⁵⁾	1994	L3 vertebral body	Fatigue, Anorexia	Tumor resection	Continued high calcium levels	N/A
Jenkins PJ ⁶⁾	1997	T1 vertebral body	Thirst, Pollakiuria	Tumor resection	Good	6 months
Zhong-ling Q ³⁾	2013	L4 vertebral body	Muscle pain, Pollakiuria	Percutaneous vertebroplasty	Good	9 months
Yifei M ²⁾	2014	L4 vertebral body	Nausea	Percutaneous vertebroplasty Radiofrequency ablation	Good	N/A
Ruan M ⁴⁾	2014	L4 vertebral body	Nausea Lower-extremity weakness	Percutaneous vertebroplasty Radiofrequency ablation	Good	N/A
Kaitlyn AB ⁷⁾	2019	C4 vertebral body	Neck and upper-extremity pain	Tumor resection	Good	N/A
Katherine MY ⁸⁾	2023	C4 vertebral body	Neck and upper-extremity pain	Tumor resection Radiotherapy	Good	15 months
Self-case	2024	C7 lamina	None (high calcium levels)	Tumor resection Radiotherapy	Good	2 years

Good: After surgery, the control of calcium levels improved.

N/A: Not Available. Not mentioned in the paper.

Ethical Approval: This study was approved by the ethics committee of Keio University School of Medicine (IRB 20110142).

Informed Consent: Informed consent for publication was obtained by the participant in this study.

References

1. Alberti A, Smussi D, Zamparini M, et al. Treatment and outcome of metastatic parathyroid carcinoma: a systematic review and pooled analysis of published cases. *Front Oncol.* 2022;12:997009.
2. Mo Y, Zhou J, Bao Y, et al. Unexpected vertebral metastasis of parathyroid carcinoma. *Chin Med J.* 2014;127(4):800.
3. Qiu ZL, Wu CG, Zhu RS, et al. Unusual case of solitary functioning bone metastasis from a "parathyroid adenoma": imagiologic diagnosis and treatment with percutaneous vertebroplasty-case report and literature review. *J Clin Endocrinol Metab.* 2013;98(9):3555-61.
4. Ruan M, Shen Y, Zhang H, et al. Bone metastasis from parathyroid carcinoma non-avid for 99mTc-MIBI, 99mTc-MDP, and 18F-FDG. *Nucl Med Radiat Ther.* 2014;5(1):165-8.
5. Koyano H, Shishiba Y, Shimizu T, et al. Successful treatment by

surgical removal of bone metastasis producing PTH: new approach to the management of metastatic parathyroid carcinoma. *Intern Med.* 1994;33(11):697-702.

6. Jenkins PJ, Satta MA, Simmgren M, et al. Metastatic parathyroid carcinoma in the MEN2A syndrome. *Clin Endocrinol.* 1997;47(6):747-51.
7. Brooks KA, Jensen BT, Naqvi SHS, et al. Metastatic parathyroid carcinoma presenting as an osteolytic cervical T vertebral mass 5 years after parathyroidectomy. *Otolaryngol Case Rep.* 2019;10:10-2.
8. Yu KM, Trimble DJ, Conner CR, et al. Oligometastatic parathyroid carcinoma presenting as neck pain: a case report. *Ear Nose Throat J.* 2023;102(10):635-9.
9. Ueki Y, Takahashi T, Ota H, et al. Parathyroid carcinoma with brown tumor mimicking multiple skeletal metastases: a case report. *Folia Endocrinol Jpn.* 2020;37(1):55-9.
10. Goswamy J, Lei M, Simo R. Parathyroid carcinoma. *Curr Opin Otolaryngol Head Neck Surg.* 2016;24(2):155-62.

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