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CASE REPORT

Successful treatment of a diffuse type tenosynovial giant cell tumor in the thoracic spine mimicking spinal metastasis by frozen recapping laminoplasty in a patient with thyroid cancer

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Abstract

Purpose Tenosynovial giant cell tumor of the diffuse type (TGCT-D) involving the spine is rare. Its differential diagnosis includes metastatic disease; however, there have been few reports of spinal TGCT-D mimicking spinal metastasis in patients with a history of malignancy.

Methods We report on a 35-year-old woman with a history of papillary thyroid cancer who was diagnosed with TGCT-D of the thoracic spine mimicking spinal metastasis. Preoperative computed tomography (CT) revealed a 1.0 × 1.0-cm lytic bone lesion involving the left T7 vertebral lamina, pedicle, and the T6–7 facet joint; the thoracic spine lesion was markedly fluorodeoxyglucose-avid on positron-emission tomography/computed tomography (PET/CT).

Results Spinal metastasis was initially suspected given the patient's history of papillary thyroid cancer. Total excision was performed with recapping laminoplasty. The resected lamina was frozen in liquid nitrogen and used as a frozen autograft (frozen recapping laminoplasty) for spinal reconstruction with posterior instrumentation. Histological findings supported a diagnosis of TGCT-D. The patient had no evidence of local recurrence 2 years post-surgery. Bone union was achieved 3 years post-surgery.

Conclusions TGCT-D can mimic metastasis in PET/CT and should be included in the differential diagnosis if a lytic lesion affecting the posterior elements of the vertebrae involves the facet joints. CT-guided biopsy is recommended for accurate diagnosis when an occult tumor, such as TGCT, is incidentally detected on PET-CT, even in patients with a history of malignant neoplasm. Frozen recapping laminoplasty is useful for complete resection of a spinal tumor, preventing local recurrence, and preservation of the posterior spinal elements.

Keywords Frozen autograft · Tenosynovial giant cell tumor · Recapping laminoplasty · Thoracic spine · Thyroid cancer

Introduction

Tenosynovial giant cell tumor of diffuse type (TGCT-D) was originally defined as a pigmented villonodular synovitis by Jaffe et al. in 1941 [1]. According to the 2013 WHO classification of soft tissue tumors, TGCT can be subdivided into localized and diffuse forms. TGCT-D is a benign proliferative disorder that primarily occurs in the large joints of the appendicular skeleton, such as the knee and hip joints [2]. It is usually found in the young age group (20–50 years), and the distribution between men and women is almost equal [3]. TGCT-D rarely presents in the spine, particularly in the thoracic segments. Its lytic radiographic appearance and high avidity on fluorodeoxyglucose-positron emission tomography (FDG-PET) can mimic more aggressive bone lesions, including metastatic disease [4–11]. However, there have been few reports of spinal TGCT-D in patients with a history of malignant neoplasm.

Recapping T-saw laminoplasty is performed as treatment for any type of spinal tumor and provides extensive exposure for tumor removal [12–14]. In the present case, en bloc resected lamina, including the tumor, was frozen in liquid nitrogen and reused as an autograft for reconstruction. We here present a case of TGCT-D of the thoracic spine that mimicked spinal metastasis in a 35-year-old woman with a

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history of papillary thyroid cancer. The patient was successfully treated with "frozen recapping laminoplasty," which facilitates complete total resection of the tumor.

Case report

A 35-year-old woman was diagnosed with papillary thyroid cancer, and a right lobectomy was performed. One month after surgery, the patient underwent an FDG-PET study to evaluate the extent of the disease. PET/computed tomography (CT) showed an intensely FDG-avid nodule in the left upper lobe of the thyroid, with a maximum standardized uptake value (SUV_{max}) of 6.8, and an incidental FDG-avid lesion in the T7 lamina with an SUV_{max} of 10.4, suggesting metastatic disease from thyroid cancer. CT revealed a 1.0 × 1.0-cm lytic bone lesion involving the left T7 vertebral lamina, pedicle, and T6–7 facet joint (Fig. 1). A heterogeneous hypointense lesion was revealed on T1-weighted magnetic resonance imaging (MRI), and T2-weighted sequences also revealed a low-intensity lesion. Gadolinium-enhanced MRI demonstrated a uniformly enhancing mass (Fig. 2). Spinal metastasis was suspected based on the history of papillary thyroid cancer. Therefore, the patient was referred to our hospital for excisional surgery of the T7 spinal tumor. The patient had no neurological deficits or back pain. Her serum thyroglobulin level was 14.0 ng/mL (reference range 1.4–78.0 ng/mL). We also suspected spinal metastasis despite a thyroglobulin-negative status, and planned total excision of the tumor with frozen recapping laminoplasty using liquid nitrogen and spinal instrumentation.

The surgery was performed via a posterior approach. In the first step, en bloc resection of the left T6 spinous process and left inferior articular process, including the tumor, was performed. Thereafter, en bloc resection of the posterior element of T7, including the tumor at the left superior articular process, was performed via pediculotomy on the side of the tumor as well as cutting from the pedicle to the transverse process on the opposite side of the tumor, using a flexible multifilament thread-wire saw (T-saw; Promedical Co, LTD, Kanazawa, Japan) [12]. En bloc laminectomy provided extensive exposure for tumor removal, and piecemeal total resection of the residual tumor was performed at the left T6/7 foramen. After en bloc laminectomy, the tumor and soft tissue were curetted away from the resected lamina. Then, the resected lamina was frozen with liquid nitrogen for 20 min and then used as a frozen autograft (frozen recapping

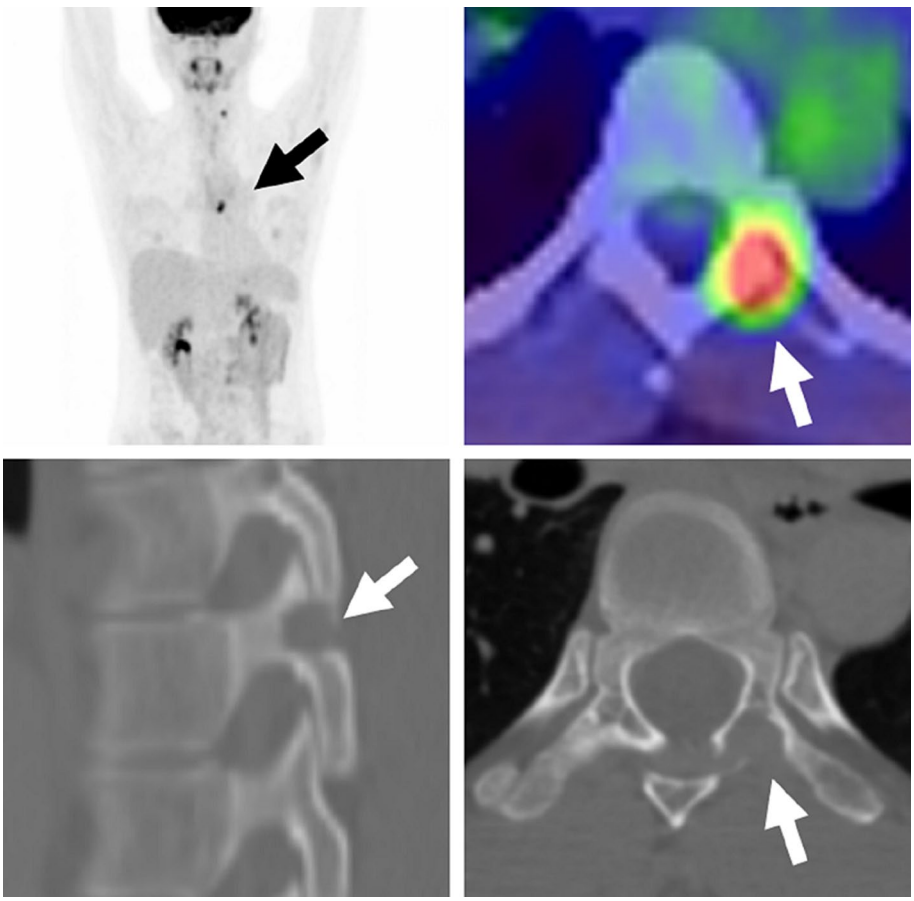

Fig. 1 Preoperative positron-emission tomography-computed tomography (CT) image and plain CT image of the thoracic spine. A positron-emission tomography-CT image showing high fluorodeoxyglucose accumulation in the left side of the T7 lamina (maximum standard update value = 10.4), and a plain CT image showing a 1-cm osteolytic tumor involving the left T7 vertebral lamina, pedicle, and T6–7 facet joint (black and white arrows)

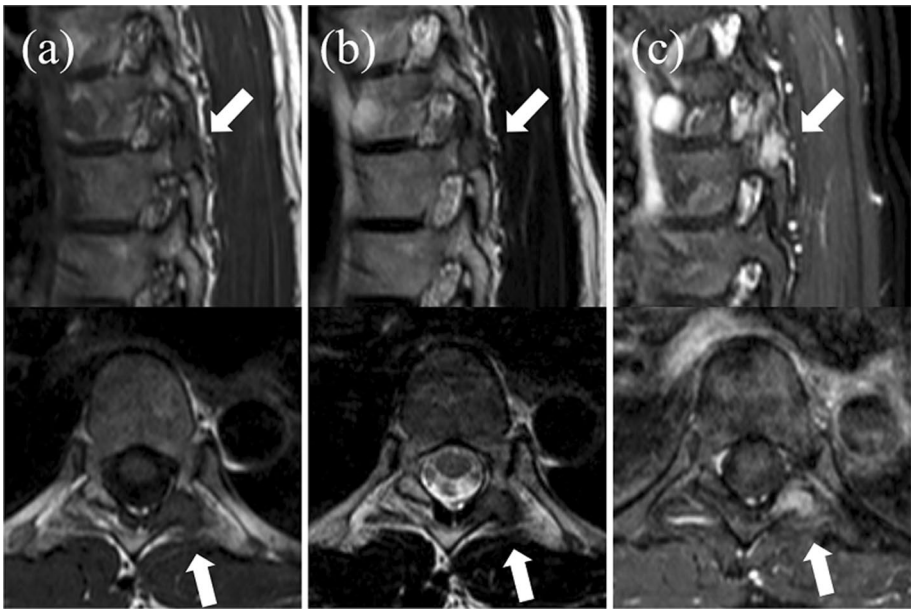

Fig. 2 Preoperative magnetic resonance imaging of the thoracic spine. T1- and T2-weighted images (**a**, **b**, respectively) showing low signals. Tumor enhancement was observed with gadolinium contrast (**c**). The white arrows indicate the tumor

laminoplasty) for spinal reconstruction with posterior instrumentation (Figs. 3, 4).

Pathological analysis revealed synovial-type tissue with a fibrohistiocytic reaction (Fig. 5). Small mononuclear cells showing spindle- to oval-shaped nuclei were diffusely distributed and mixed with several multinucleated giant cells, which is characteristic of TGCT-D. There was no evidence of malignancy. Immunohistochemistry showed that the majority of cells were vimentin- and CD68-positive, but negative for CD1a, CD3, CD20, CD138, and S-100.

There were no perioperative complications. CT-guided biopsy for the left upper lobe of the thyroid was performed after spine surgery and tumor cells were not detected. The left upper lobe of the thyroid was followed without therapy at final follow-up. MRI was performed 2 years after the surgery and revealed no evidence of local recurrence (Fig. 6). One year after the surgery, bone union had not yet been completely achieved; however, 3 years after the surgery, bone union was complete. At the 3-year post-surgery follow-up, radiography and CT demonstrated that the reconstructed spine was well maintained (Fig. 7).

Discussion

Tenosynovial giant cell tumor of the diffuse type is a slowly progressing, benign, but locally aggressive lesion of unknown etiology. The annual incidence of TGCT-D is estimated to be 1.8 per million individuals [15], and the knee joint is the most commonly afflicted region (up to 53.4% of cases) [16]. Spinal TGCT-D is very rare, particularly thoracic TGCT-D. The cervical spine was most frequently involved (50–73%), with infrequent involvement of the lumbosacral (20–25%) and thoracic spine (7–25%) [17].

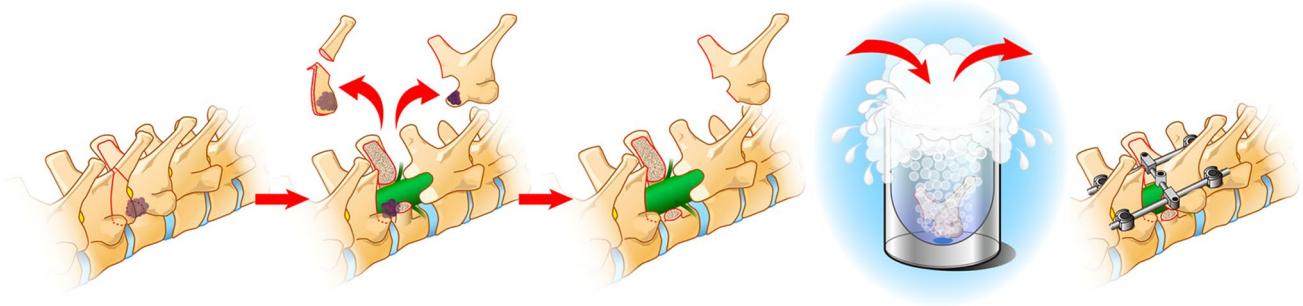

Fig. 3 Diagram illustrating frozen recapping laminoplasty. En bloc resection of the posterior element of T7 was performed by pediculotomy on the tumor side and by cutting from the pedicle to the transverse process on the opposite side of the tumor using a flexible multifilament thread-wire saw. After en bloc laminectomy, the tumor and soft tissue were curetted away from the resected lamina. Then, the resected lamina was frozen in liquid nitrogen for 20 min and used as a frozen autograft for spinal reconstruction with posterior instrumentation

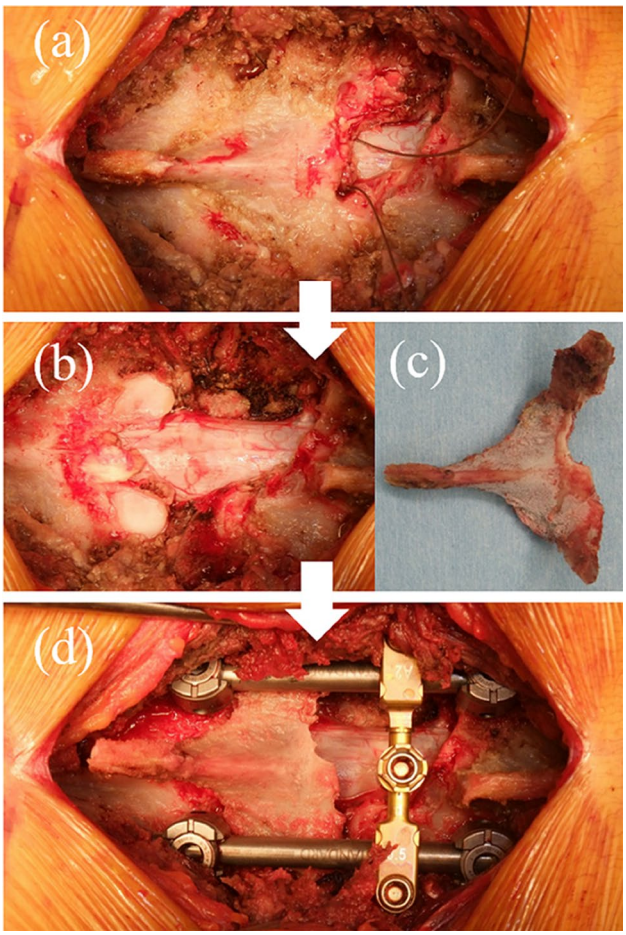

Fig. 4 Intraoperative photographs. En bloc laminectomy provided extensive exposure for tumor removal, and piecemeal total resection of the residual tumor was performed at the left T6–7 foramen (**a**, **b**). The resected lamina (**c**) was frozen in liquid nitrogen for 20 min, and used as a frozen autograft for spinal reconstruction with posterior instrumentation (**d**)

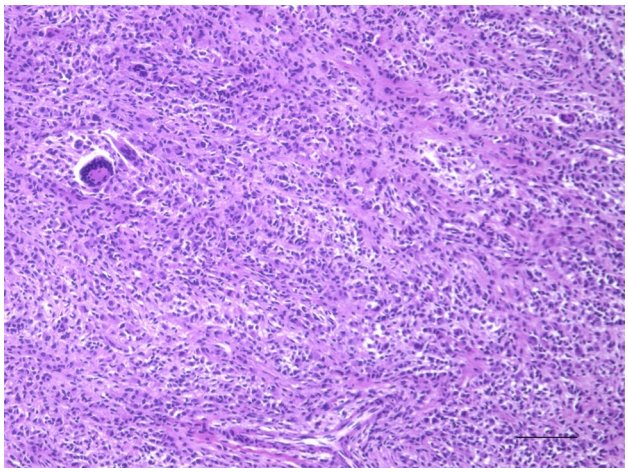

Fig. 5 Pathological findings of the resected tumor (hematoxylin and eosin staining). Small mononuclear cells showing spindle- to oval-shaped nuclei were diffusely distributed and mixed with several multinucleated giant cells, which is characteristic of tenosynovial giant cell tumor of the diffuse type (TGCT-D). Scale bar corresponds to 100 μm

TGCT-D is typically benign, but shows aggressive imaging features, and can mimic malignant tumours. However, there have been few reports of TGCT-D in patients with a history of a malignant neoplasm [4–11]. Mahmood and de Llano reported TGCT involving the right knee joint in a 47-year-old man with a history of malignant melanoma after wide local excision of the primary lesion from the left chest wall [4]. Chang et al. reported a TGCT-D involving the T9 vertebra in a 33-year-old woman with a history of papillary thyroid cancer [8]. Lavrador et al. reported the case of a 64-year-old man with C1–C2 TGCT-D and a history of renal cell carcinoma [11]. One reason for the rarity of this condition may be that the mean age at onset is low, particularly for thoracic TGCT-D. Roguski et al. reported that the mean age of patients diagnosed with thoracic TGCT-D in their cohort was 22.8 years, which was significantly lower than for those diagnosed with cervical and lumbar TGCT-D (42.4 and 48.6 years, respectively; $p = 0.0001$) [18].

It is difficult to distinguish spinal TGCT-D from spinal metastases because of their radiological similarities, including an osteolytic lesion on CT and high FDG-avidity in both metastatic and TGCT-D lesions on FDG-PET [4–11]. In the report by Mahmood and de Llano, the patient was referred for an F-18 FDG-PET examination for evaluation of the extent of malignant melanoma. PET/CT showed an incidental FDG-avid lesion in the right knee joint, with an SUV_{max} of 10.2, mimicking metastatic melanoma [4]. In the report by Chang et al., the patient was referred for PET/CT as part of a routine follow-up 4 years after thyroidectomy for papillary thyroid cancer, which revealed an incidental focal hypermetabolic lesion with a maximum SUV of 7.1; the CT component images revealed a small osteolytic lesion in the left facet of the T9 vertebra mimicking spinal metastasis [8]. The SUV_{max} values for TGCT-D are variable, ranging from 4.4 to 25.0 [9, 19]; Chang et al. argued that it would not be possible to differentiate between TGCT-D and other benign or malignant bone lesions using SUV alone, and concluded that MRI is more useful for the evaluation of musculoskeletal lesions [8]. In the present case, CT findings revealed a lytic bone lesion, and FDG-PET/CT imaging demonstrated that the thoracic spine lesion was markedly FDG-avid, mimicking spinal metastasis.

MRI findings of TGCT lesions typically demonstrate intermediate and/or low signal intensity on T2-weighted imaging, reflecting hemosiderin deposition [20], whereas most osteolytic spinal metastases show high signal intensity on T2-weighted imaging [21]; such findings on MRI may be

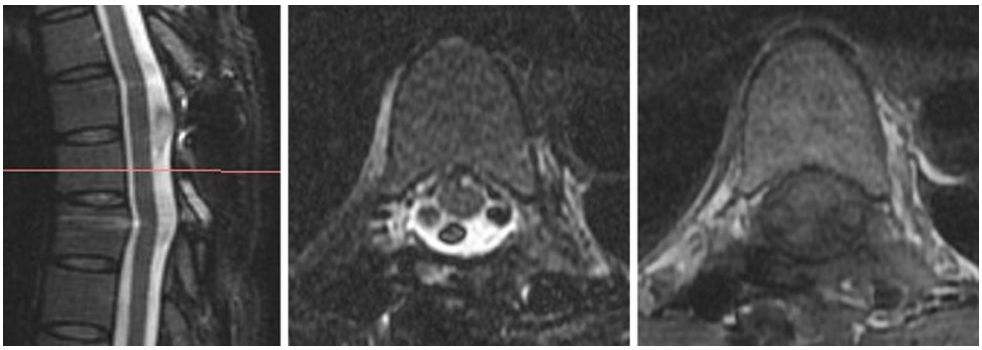

Fig. 6 Postoperative magnetic resonance imaging (MRI) obtained 2 years after the surgery. At the 2-year post-surgery follow-up, MRI revealed no evidence of local recurrence

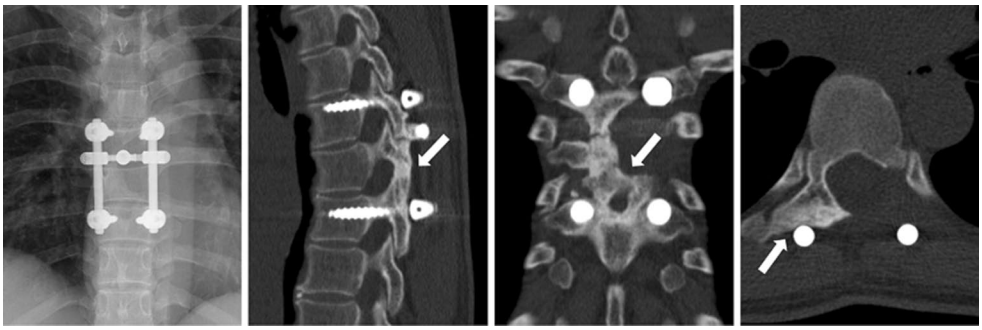

Fig. 7 Postoperative radiography and computed tomography images obtained 3 years after spinal surgery. At the 3-year post-surgery follow-up, there was no loosening of the posterior instrumentation. Bone union after frozen recapping laminoplasty was obtained (white arrows)

useful for differential diagnosis. However, MRI findings are not always characteristic. Parmar et al. reported an unusual case of TGCT-D involving the cervical spine, which was hyperintense on T2-weighted imaging [22]. The authors proposed that TGCT-D may be variable in appearance, depending on the composition of the lesion and the relative proportion of hemosiderin, lipid, fibrous tissue, cyst formation, and cellular elements.

Tumor localization and extension are the most important characteristic radiological findings that can be used to differentiate a metastatic tumor from spinal TGCT-D. Motamedi et al. presented 15 spinal TGCT-D cases in which 93% involved posterior spine elements and 67% involved the facet joint [23]. The authors concluded that, in small lesions (< 3 cm in all dimensions), a facet origin can be identified on CT or MRI, as the lesion is often confined to this joint. In contrast, spinal metastasis is common in the vertebral body and extends to the posterior elements through the pedicle. The rate of spinal metastasis in the posterior elements alone is low, at 19.5% [21]. Metastatic tumors generally extend within the same compartment of the spine; therefore, the observation that the tumor in our patient invaded the adjacent vertebra through the facet joint was atypical of spinal metastasis, especially as the tumor was small.

The most common site of bone metastasis in patients with thyroid cancer is the spine, accounting for approximately half of all such patients [24–26]. Another retrospective analysis of 202 cases of thyroid spinal metastasis revealed that almost half of all patients with single-site spinal metastasis at the time of presentation did not exhibit any other distant metastases [27]. FDG-PET can detect occult metastasis of thyroid cancer. Therefore, we originally misdiagnosed the tumor as occult spinal metastasis of thyroid cancer, without considering the possibility of TGCT. However, TGCT has also previously unexpectedly been detected with FDG-PET in patients with a history of malignant neoplasm [4–11]. Therefore, to determine the management of solitary spinal tumor, such as in the present case, CT-guided biopsy is recommended for accurate diagnosis when an occult tumor, such as TGCT, is incidentally detected on PET-CT, even in patients with a history of malignant neoplasm.

Tomita and Kawahara et al. reported successful results with recapping T-saw laminoplasty [12–14]; this procedure allows wide exposure and complete en-bloc resection of the tumor. Recapping T-saw laminoplasty can be performed to treat any type of spinal tumor. Tsuchiya et al. reported that reconstruction using frozen tumor-bearing bone is a safe and effective method [28]; no local recurrences from

tumor-bearing autografts have been reported. Using frozen autografts has various advantages, including low cost, maintenance of osteoinductive and osteoconductive properties, good fit between graft and host bone, and no transmission of disease or immunological rejection [29–31]. Total resection is the recommended first-line therapy for both TGCT-D of the spine and thyroid spinal metastasis [32–35]. Therefore, we suggest that the combination of recapping laminoplasty and reconstruction using a frozen tumor-bearing vertebra may offer an ideal surgical option for complete resection of the spinal tumor, as shown in our case, in terms of prevention of local recurrence and preservation of the posterior spinal elements.

Conclusions

We reported a rare case of thoracic TGCT-D mimicking spinal metastasis in a patient with thyroid cancer. It is difficult to differentiate spinal TGCT-D from a metastatic tumor because of the presence of a destructive lesion on CT and high FDG avidity on PET. TGCT-D should be included in the differential diagnosis of osteodestructive spinal lesions, particularly if patients are young and the tumor originates in the facet joint. To determine the management of a solitary spinal tumor, CT-guided biopsy is recommended when an occult tumor is incidentally detected on PET-CT, even in patients with a history of malignant neoplasm, such as the present case. The combination of recapping laminoplasty and reconstruction using a frozen tumor-bearing vertebra may offer an ideal surgical option for the complete resection of spinal tumors, as shown in the present case.

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Compliance with ethical standards

Conflict of interest N. Yonezawa, H. Murakami, S. Kato, H. Hayashi, and H. Tsuchiya declare that they have no conflict of interest. This study was not funded by any external source.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from the patient described in the study.

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