

A case of ectopic cervical thymoma with myasthenia gravis mimicking a parathyroid tumour

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Abstract

Reports of cervical thymoma with myasthenia gravis are rare. In addition, ^{99m}Tc-MIBI (methoxyisobutylisonitrile:sestamibi) scintigraphy is a useful diagnostic examination for enlarged parathyroid tumours; however, there are a few reports of its accumulation in thymoma. Among them, there are no reports of cervical thymomas with ^{99m}Tc-MIBI accumulation complicated by myasthenia gravis. In this study, we performed surgery on a patient with preoperative myasthenic crisis accompanied by a cervical thymoma and a parathyroid tumour. Preoperatively, the cervical mass was determined to be a parathyroid tumour and was complicated by myasthenia gravis without thymic tumour. However, a pathological examination revealed that the cervical tumour with ^{99m}Tc-MIBI accumulation was a Type B2 thymoma, and a parathyroid tumour was identified in the vicinity. We report a very rare case in which symptoms improved with surgery.

KEYWORDS

^{99m}Tc-MIBI accumulation, cervical thymoma, myasthenia gravis, parathyroid tumour

INTRODUCTION

^{99m}TcMIBI scintigraphy was performed for a cervical mass with hypercalcemia to differentiate it from a parathyroid tumour. Based on the results, we considered the cervical tumour to be a parathyroid tumour, which was also complicated by myasthenia gravis due to generalized muscle weakness and high levels of anti-acetylcholine receptor antibodies. Since there was no thymoma in the mediastinum, we considered myasthenia gravis without thymoma. However, the pathology results showed a cervical thymoma with positive ^{99m}TcMIBI scintigraphy. In addition, a small parathyroid tumour that was not noted on the images was identified near the thymoma. We report this case because the patient developed myasthenic crisis in the perioperative period and was difficult to diagnose and treat.

CASE REPORT

The patient was a 61-year-old woman with no specific medical history. She was aware of the feeling of fatigue and drooping eyelids, and was examined closely and diagnosed with myasthenia gravis. During the course of the disease, the patient's swallowing function deteriorated and she was admitted to the hospital with a diagnosis of myasthenic crisis. Peripheral blood smear test results were normal; however, her serum calcium (10.5 mg/dL), serum albumin (3.9 g/dL), intact parathyroid hormone (PTH; 111 pg/mL), and anti-acetylcholine receptor antibody (301 nmol/L) values were high. A chest X-ray showed no obvious abnormalities in the lung fields; however, the trachea was shifted to the right due to the mass (Figure 1a). Contrast-enhanced computed tomography (CT) showed a lobulated cystic lesion in the neck and a full tumour of 4.5 cm in size with smooth margins and a

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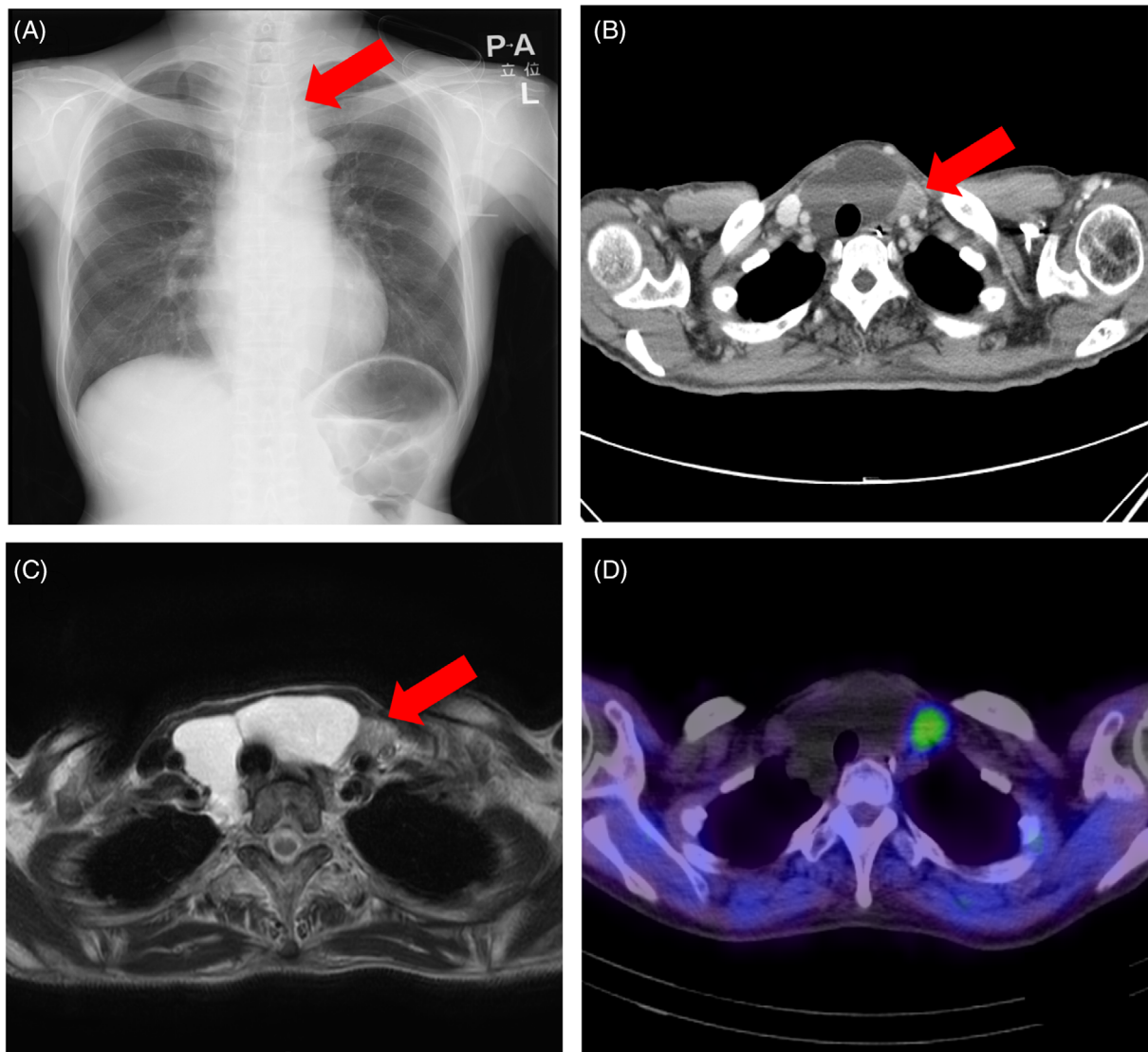


FIGURE 1 (a) Chest X-ray showed no obvious abnormalities in the lung fields; however, the trachea was shifted to the right due to the mass. (b) The trachea was compressed to the right by the tumour. There was no narrowing of the tracheal lumen. (c) Simple magnetic resonance imaging (MRI) showed a lobular cystic lesion and a substantial tumour of 4.5 cm in size, both with clear borders and no tendency to invade the surrounding area. (d) $^{99m}\text{TcMIBI}$ scintigraphy showed enhanced accumulation consistent with the tumour

septal wall between them. The trachea was compressed to the right by the tumour; however, there was no narrowing of the tracheal lumen (Figure 1b). Simple magnetic resonance imaging (MRI) showed a lobular cystic lesion and a 4.5 cm-sized substantial tumour, both with clear borders and no tendency to invade the surrounding organs (Figure 1c). $^{99m}\text{TcMIBI}$ scintigraphy showed enhanced accumulation consistent with the tumour (Figure 1d).

After admission, treatment with anticholinesterase agents and steroids was introduced and tube feeding was started. At the same time, she underwent immunoadsorption plasmapheresis (IAPP) a total of six times before surgery. Her muscle weakness tended to improve, and surgery was performed 53 days after admission.

Thoracoscopic extended thymectomy was performed in the supine position and the tumour was removed by a cervical approach. On the second postoperative day, respiratory distress appeared and an arterial blood gas analysis showed carbon dioxide retention. The patient was judged to have postoperative crisis and non-invasive positive pressure ventilation (NPPV) was started. She could be weaned off NPPV after receiving IAPP twice in the subsequent course and was transferred to the hospital on postoperative day 52. Serum calcium and intact parathyroid hormone (PTH) were also within reference values at discharge.

Histopathology revealed numerous small lymphocytes and round nucleated epithelial-like cells within the 4.5-cm tumour, leading to a diagnosis of type B2 thymoma

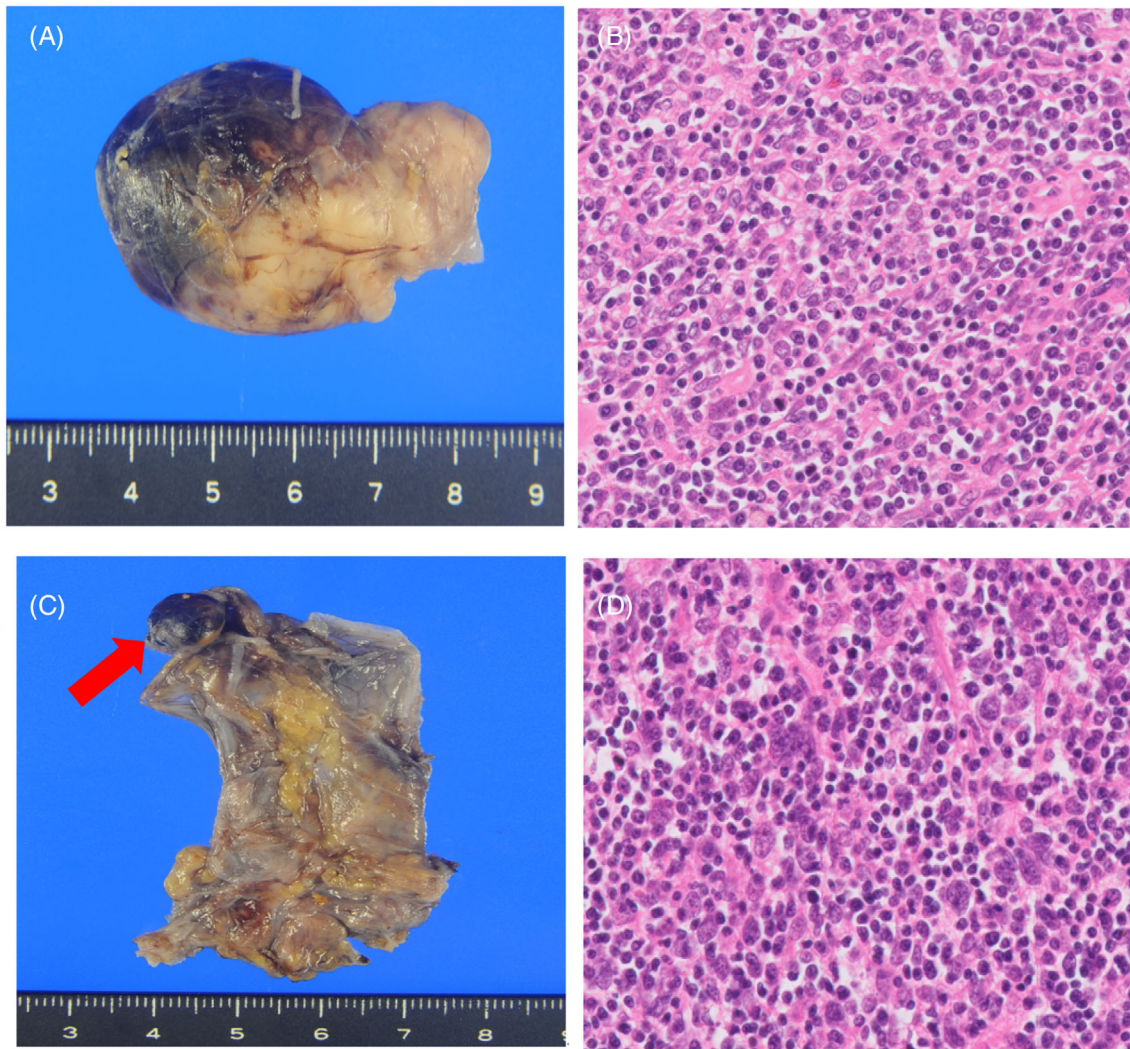


FIGURE 2 (a), (b) Histopathology revealed numerous small lymphocytes and round nucleated epithelial-like cells within the 4.5-cm tumour, leading to a diagnosis of type B2 thymoma. (c), (d) The 1-cm tumour attached to the thymic cyst wall was a sheet-like growth of main cell-like tumour cells with a mildly enlarged nucleus, and was diagnosed as parathyroid adenoma

(Figure 2a,2b). The 1-cm tumour attached to the cyst wall was a sheet-like growth of main cell-like tumour cells with a mildly enlarged nucleus, and was diagnosed with parathyroid adenoma (Figure 2c, d). The cystic lesion was diagnosed as a thymic cyst (Figure 2c).

DISCUSSION

^{99m}TcMIBI scintigraphy is the standard method for the localization of enlarged parathyroid glands. The sensitivity of ^{99m}TcMIBI scintigraphy for preoperative localization of lesions in hyperparathyroidism is reported to be 72%–84.9%.¹ In addition, MIBI may physiologically accumulate in the thyroid gland, salivary glands, nasal mucosa, oral cavity, myocardium, liver, and gastrointestinal tract, but not in the normal parathyroid gland with a weight of 30–40 mg.² In the present case, ^{99m}TcMIBI scintigraphy showed

accumulation that was consistent with a 4.5 cm substantial tumour, and pathology showed that the tumour was a Type B2 thymoma. The mechanism of accumulation in the thymoma has not been clearly elucidated, although it is believed to be closely related to the thymoma, since the inferior parathyroid organs arise from the third pharyngeal cone adjacent to the thymic organs during the embryonic period.³ In addition, most thymomas occur in the anterior mediastinum, and about 4% occur in the neck, middle or posterior mediastinum.⁴ Among them, reports of cases complicated by myasthenia gravis are quite rare. The patient had preoperative myasthenia gravis crisis, suggesting that the cervical mass may have been thymoma; however, the high serum calcium value and intact PTH value, and the accumulation on ^{99m}TcMIBI scintigraphy⁵ made it difficult to distinguish from a parathyroid tumour. Extended thymectomy and neck tumour excision revealed a parathyroid tumour of 1 cm in size in the vicinity of the removed thymoma. The symptoms

of myasthenia gravis gradually lessened after the surgical removal of the tumour.

We experienced and reported an extremely rare case of cervical thymoma with $^{99m}\text{TcMIBI}$ scintigraphic accumulation associated with myasthenia gravis.

AUTHOR CONTRIBUTIONS

Go Kamimura is the first author and Kazuhiro Ueda is the corresponding author of this manuscript. Aya Takeda, Ryo Miyata, Masaya Aoki, and Toshiyuki Nagata participated in the operation of this case. Masami Sato supervised the operation and the editing of the manuscript. Go Kamimura and Kazuhiro Ueda drafted the manuscript, and all authors read and approved the final manuscript.

CONFLICT OF INTEREST

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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