Int Canc Conf J (2016) 5:158–161
DOI 10.1007/s13691-016-0249-7

CASE REPORT

Metachronous multiple thymoma with different clinical behavior and pathological findings

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Received: 18 February 2016 / Accepted: 29 March 2016 / Published online: 16 April 2016

Abstract A 70-year-old woman presented with a nodule in the left hilum on a chest radiograph 3 years before. Another mass emerged caudal to the initial nodule and was diagnosed as thymoma. A surgical specimen revealed two components: an encapsulated rostral nodule and a caudal mass invading the left lung. Histological findings showed that the rostral nodule was a stage 1 type B2 thymoma, whereas the caudal mass was a stage 3 type B3 thymoma. Based on the differences in biological behavior and histological findings, we concluded that these tumors derived from multicentric origin.

Keywords Metachronous · Multiple · Thymoma · Multicentric

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Abbreviations

MG	Myasthenia gravis
CT	Computed tomography
WHO	World Health Organization

Introduction

Multiple thymoma is a very rare entity accounting for 1–2 % of all thymomas [1, 2]. There are two hypotheses about their origin: deriving from multicentric development or intra-thymic metastasis. We herein report a case that supports multicentric origin based on the clinical course and histological findings.

Case report

A 70-year-old woman without myasthenia gravis (MG) presented with a nodule in the left hilum on a chest radiograph 3 years before (Fig. 1a). Because it was calcified with a clear margin, suggesting a benign lesion, annual follow-up was chosen. She developed another mass, which emerged caudal to the initial nodule, and she was referred to our hospital for further investigation (Fig. 1b). Chest computed tomography (CT) revealed an encapsulated rostral nodule with calcification, as well as a caudal mass adhered to the left lung and pericardium (Fig. 2). CT-guided needle biopsy obtained from the caudal mass revealed the histological diagnosis as thymoma, and she underwent thymomectomy with wedge resections of the left lung and the pericardium by median sternotomy. The surgical specimen was composed of two lesions, with the rostral

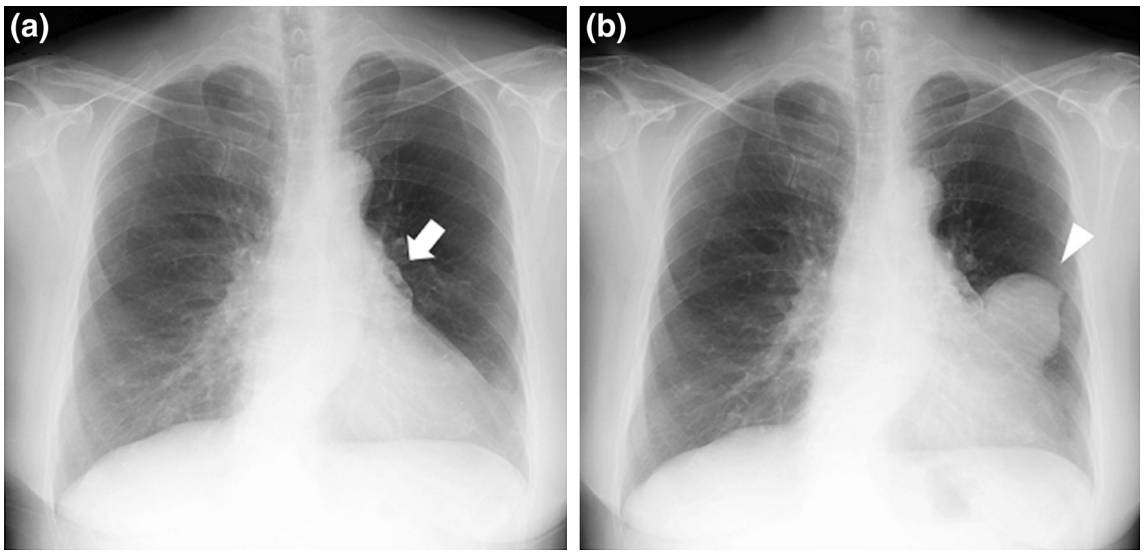

Fig. 1 **a** Chest radiograph showed a calcified nodule in the left pulmonary hilum (*white arrow*). **b** Another mass emerged caudal to the initial nodule 3 years thereafter (*white arrow head*)

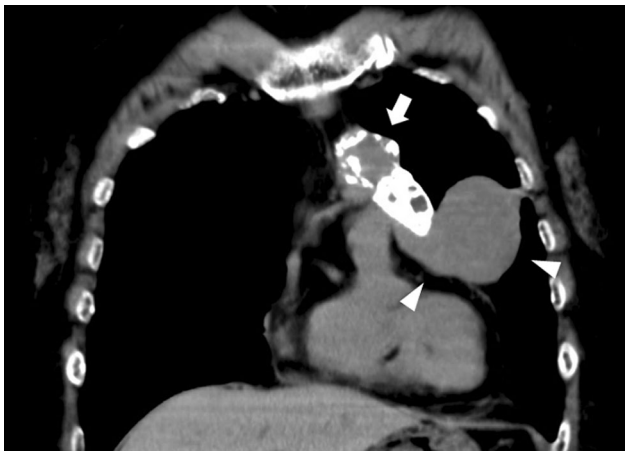

Fig. 2 Chest CT revealed a calcified nodule at the anterior mediastinum (*white arrow*) and a caudal mass adhered to the left lung and pericardium (*white arrowheads*)

nodule clearly distinguished from the surrounding tissue and the caudal mass strictly adhered to the lung (Fig. 3). Histological and immunohistochemical examination of the rostral nodule revealed a type B2 thymoma according to the World Health Organization (WHO) classification, shown to be stage 1 according to the Masaoka classification (Fig. 4a); on the other hand, the caudal mass was a type B3 thymoma, shown to be stage 3 due to microscopic invasion of the lung (Fig. 4b). The thickened connective tissue separated the two lesions histologically. Because of the absence of more aggressive rapidly growing caudal mass on a chest radiograph 3 years before we considered that they emerged metachronously. As the tumors had different histologies, clinical features and no histological continuity, we concluded that they derived from multicentric origin. Postoperative course was uneventful. She was discharged on the fourth postoperative day and had no sign of recurrence at 1 year.

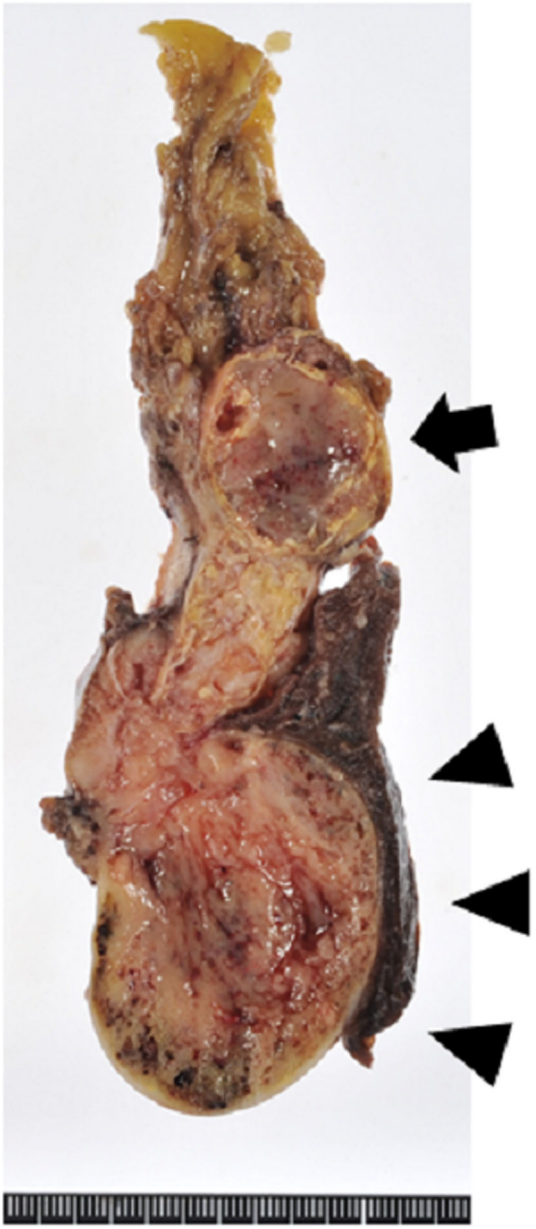

Fig. 3 The cut surface of the surgical specimen revealed two distinct components separated by thickened connective tissue, the encapsulated rostral nodule (*black arrow*) and the caudal mass invading the lung (*black arrowheads*)

Discussion

Although thymoma is the most common tumor of the anterior mediastinum, multiple developments are uncommon [3–6]. Moreover, metachronous onset, which occurred over 3 years in the present case, is extremely rare; most reported cases have involved synchronous onset. There are two hypotheses as to the origin of multiple thymoma: deriving from multicentric development or intra-thymic metastasis. In the case of different histological subtypes, the multicentric hypothesis seems more suitable [3, 7]. Intra-thymic metastasis, however, cannot be excluded if the tumors have the same characteristics [8]. The two lesions in the present case showed different clinical and pathological features. The initial rostral lesion had been stable in size for 3 years and was shown to be an encapsulated type B2 thymoma. In contrast, the caudal lesion emerged rapidly and was shown to be an invasive type B3 thymoma. Therefore, we concluded that these tumors derived from multicentric origin.

There is debate as to the optimal mode of resection of a thymoma. While recent guidelines recommend thymothymomectomy, thymomectomy might be a feasible alternative in patients without MG [9, 10]. However, multiple thymoma can develop metachronously, as shown in the present case; thus, thymothymomectomy may be required to prevent another primary or metastatic lesion in the remnant thymus. Because the patient in the present case underwent a thymomectomy, she is being carefully observed at an outpatient clinic.

Conclusion

We encountered a case of metachronous multiple thymoma. Based on the differences in biological behavior and histological findings, we concluded that these tumors derived from multicentric origin. Thymothymomectomy might be required even for stage 1 thymoma because of the possibility of metachronous multicentric development.

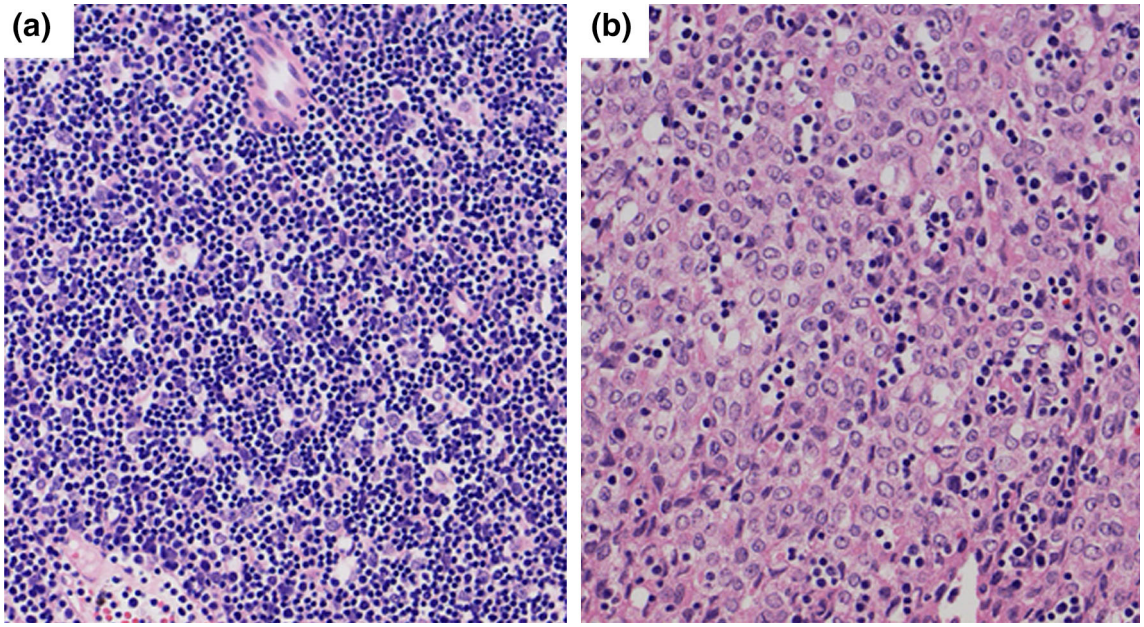

Fig. 4 **a** The rostral tumor was diagnosed as type B2 thymoma with rich immature lymphocytes. **b** The caudal tumor was diagnosed as type B3 thymoma with more epithelial cells than lymphocytes (hematoxylin and eosin stain, original magnification ×200)

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Research involving human participants and/or animals This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent We routinely obtain general consent from every patient prior to surgery before using his or her clinical data. Written informed consent was not obtained from the patient for publication of this case report because this report is merely a retrospective case report without additional invasive examinations or treatments for a study.

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