

CASE REPORT

Invasive thymoma with multiple ring calcifications and osseous metaplasia: A case report

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Abstract

An 8.0 × 7.0 × 3.0 cm calcified anterior mediastinal mass was found in a 57-year-old man during a regular health checkup. The tumor had invaded the pericardium and phrenic nerve. The Masaoka–Koga classification was stage III. Multiple ring calcifications were present in the gross feature, and osseous metaplasia was observed in the histologic examination. World Health Organization histologic classification of the tumor was type B2. The patient is currently undergoing chemotherapy and radiation therapy to prevent tumor recurrence. To our knowledge, this is the first case of multiple ring calcifications and osseous metaplasia in invasive thymoma.

Introduction

Thymoma is the most common anterior mediastinal tumor. It is often difficult to distinguish between an invasive and an encapsulated thymoma.^{1,2} Calcification is detected in 10–41% of thymomas and in approximately 54% of invasive thymomas. Although various forms of calcification are observed in thymomas, the most common type is small foci of calcification. Thymomas with massive calcification are rare.^{3,4} We describe a case of multiple ring calcifications and osseous metaplasia in a thymoma.

Case report

An abnormal shadow with anterior mediastinal calcification and an elevated left hemidiaphragm was incidentally observed in a 57-year-old man who underwent chest radiography during a periodic health examination (Fig 1). Chest computed tomography (CT) revealed an anterior mediastinal mass with multiple ring calcifications (Fig 2). He denied a relevant medical history and symptoms such as muscle weakness or red blood cell hyperplasia. Physical examination revealed no abnormal findings and no



Figure 1 Chest X-ray showing mediastinal calcification.

lymphadenopathy. Routine laboratory parameters were within reference ranges. He reported no history of other operations.

Based on a high index of clinical suspicion for a non-invasive thymoma or teratoma, we decided to perform surgery for accurate diagnosis and treatment. After induction of general anesthesia, an extended total thymectomy and concomitant pericardial resection were performed via a median sternotomy. Using blunt and sharp dissection, the gland was freed from the pericardium and the adjacent mediastinal pleura. Although the tumor was observed to have invaded the pericardium and the left phrenic nerve, the extent of tumor invasion was unclear. A cardiovascular surgeon was consulted, and the invaded pericardium and the left phrenic nerve were carefully resected. The tumor had not invaded the heart, and the remaining thymic tissue was removed.

The resected tumor measured $8.0 \times 7.0 \times 3.0$ cm in size (Fig 3a) and was observed to invade the pericardium directly. In gross sections, the tumor appeared to be a solid mass separated by multiple rings of calcification, and the surgical stage was identified as Masaoka–Koga stage III.

Histopathological examination revealed that the tumor was encapsulated by a calcified and ossified layer with invasion of the pericardium and pericardial fat tissue (Fig 3a,b). A high-power view showed epithelioid cells intermingled with lymphocytes and perivascular lymphocytic infiltration (Fig 3c,d). Based on World Health Organization histopathological classification, the patient was diagnosed with an invasive thymoma type B2.

The patient's postoperative course was uneventful, and he was discharged on postoperative day 12. The patient is currently undergoing chemotherapy and radiation therapy to prevent tumor recurrence.

The human research ethics committee of Gyeongsang National University Hospital provided a waiver as approval is not necessary for single-case reports. The patient provided written informed consent for the publication of clinical details and images.

Discussion

Several studies have reported the occurrence of calcified thymomas. Calcification alone does not characterize an invasive thymoma, as various forms of calcification are known to occur. The most common form of calcification in a thymoma is the small foci-type,^{3–6} and thymomas with single ring-type calcifications are rarely reported.⁷ Moreover, to our knowledge, multiple ring calcifications have never been reported in thymomas. Thymomas with osseous metaplasia were first reported by Funai *et al.*⁵ A few cases of osseous metaplasia in thymomas have been reported thereafter⁸ however, thymomas with both multiple ring-type calcifications and osseous metaplasia have never been reported. Intramural ossification and

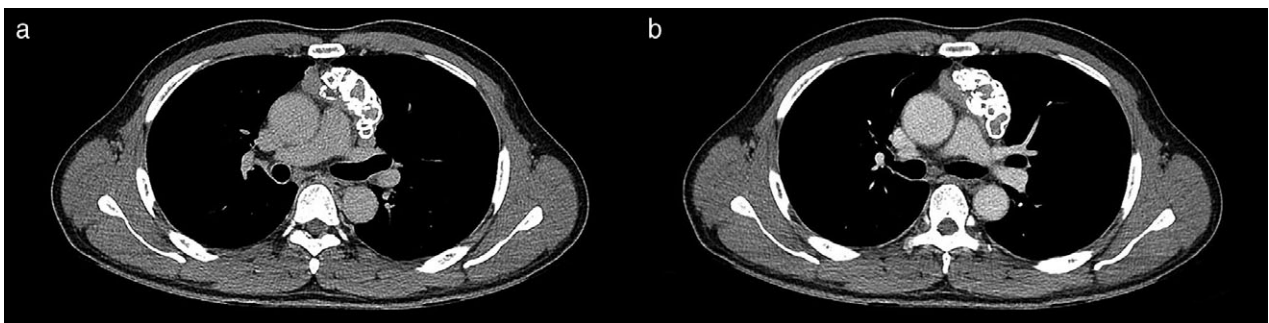


Figure 2 (a) Unenhanced chest computed tomography (CT) scan showing an anterior mediastinal mass with multiple ring calcifications. (b) Contrast-enhanced chest CT scan obtained in the delayed phase showing mild enhancement of the soft tissue component extending into the pericardial fat tissue.

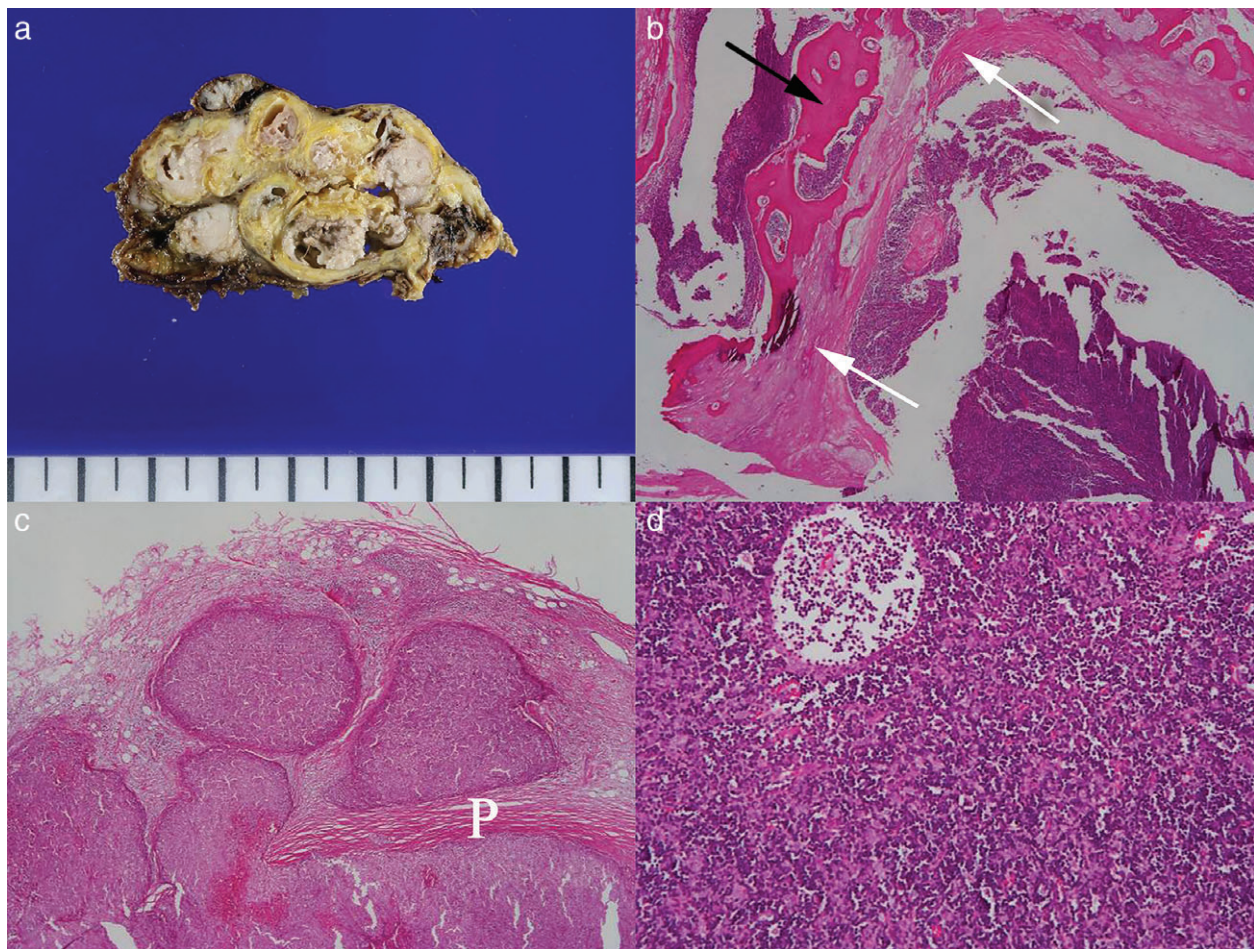


Figure 3 (a) Gross appearance of the tumor: the resected tumor appeared to be a solid mass separated by multiple foci of calcification, demonstrating a honeycomb appearance. (b) Microscopic features: the tumor appeared to be encapsulated by a calcified (white arrows) and ossified (black arrow) layer (hematoxylin and eosin [H&E] stain $\times 20$). (c) Tumor cells showing invasion of the pericardium (P) and pericardial fat (H&E stain $\times 20$). (d) High-power view showing epithelioid cells intermingled with lymphocytes and perivascular lymphocytic infiltration consistent with an invasive thymoma, classified as World Health Organization type B2 (H&E stain $\times 100$).

calcification have been described in neoplasms originating from various organs such as the kidneys, liver, gastrointestinal tissues, breast, thyroid, skin, and soft tissue. Bone formation within intrathoracic tumors has been reported in patients with teratomas, mesotheliomas, and lung cancer.⁵

It is difficult to distinguish between ossification and calcification radiographically. However, it is important to accurately identify and distinguish between ossification and calcification with respect to anterior mediastinal lesions to establish the differential diagnoses of teratomas and other tumors, such as a thymoma, dermoid and neurogenic tumors, and lymphadenopathy.^{5,7}

Usually the anatomical relationship between a tumor and the surrounding tissues of thymomas without calcification or ossification can easily be identified on a chest CT scan. In the present case, the high degree of attenuation of

multiple ring calcifications made it difficult to ascertain whether the tumor had invaded the surrounding tissues. Therefore, it was difficult to plan definitive surgery. We suspected that a tumor with this type of calcification would be benign; however, the tumor was observed intraoperatively to have invaded the pericardium and the phrenic nerve and showed dense adhesions to the epicardium. An extended total thymectomy and pericardial resection was completed without cardiopulmonary bypass.

The mechanism of calcification in thymoma patients occurs in the fibrous capsule of the thymoma, resulting in ring-shaped calcifications.⁹ In this case, the multilobulated cavities were different from the single cavity, and calcification occurred in the fibrous tissue of each cavity.

The highlights of this case are: (i) multiple ring calcifications and concomitant osseous metaplasia are extremely

rare in thymomas; (ii) benign and malignant tumors with this type of ring calcification cannot be distinguished; and (iii) it is necessary to consult a cardiac surgeon and remain prepared to perform a cardiopulmonary bypass before performing surgery because this kind of tumor can invade surrounding tissues, such as the pericardium, the heart, and the great vessels.

In summary, we conclude that it is difficult to ascertain the extent of invasion in patients presenting with an invasive thymoma showing multiple ring calcifications and concomitant osseous metaplasia based on preoperative CT findings. It is necessary to prepare for invasion when determining the surgical course.

Disclosure

No authors report any conflict of interest.

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