Rupture of thymoma due to recurrent tumor hemorrhage: a case report

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Background: Rupture of a thymoma is rare, and due to its rarity, the mechanism of rupture remains unclear. Here we report a case of a ruptured thymoma that ruptured due to an increase in the intratumoral pressure caused by recurrent hemorrhaging.

Case Description: A 70-year-old woman presented 2 days prior persistent right chest and shoulder pain. A chest computed tomography (CT) scan revealed the presence of a mass occupying the anterior mediastinum and a right pleural effusion. It was diagnosed as an anterior mediastinum tumor. The increase in the levels of inflammatory markers and tumor necrosis observed on CT were suggestive of infection. As the general status of the patient was stable and she initially received antibiotic medical therapy, an improvement in the inflammatory marker levels was observed with antibiotic therapy. A surgical resection was performed 10 days after admission. Median sternotomy revealed a tumor extending from the mediastinum to the right thoracic cavity. Since the adhesion was strong and tumor invasion was suspected, the tumor was completely resected by combining a partial resection of the right middle and lower lobes with the pericardium. Pathological examination revealed that the tumor was a type B2 thymoma with fibrosis, necrosis, hemosiderosis, and hemorrhaging, suggesting recurrent hemorrhaging within the tumor.

Conclusions: Based on the findings of our case, recurrent hemorrhaging within the tumor led to an increase in the intratumoral pressure and chronic inflammation and necrosis weakened the tumor wall. These changes contributed to the subsequent rupture.

Keywords: Thymoma; rupture; necrosis; hemorrhage; case report

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Introduction

A thymoma is the most common anterior mediastinal tumor in adults, accounting for 20% of all anterior mediastinal tumors (1). The tumors are usually diagnosed incidentally on chest radiographs. Rupture of a thymoma is rare, and due to its rarity, the mechanism of rupture is unclear. Here we report a case of thymoma that ruptured due to an increase in the intratumoral pressure caused by recurrent

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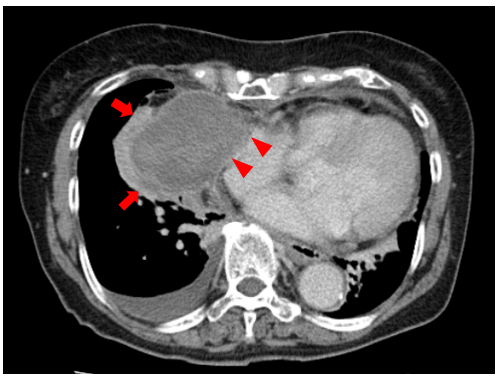

Figure 1 Chest CT on admission. Chest enhanced CT revealed the presence of an 85 mm × 60 mm mass occupying the anterior mediastinum and a right pleural effusion. The interior of the tumor was unreinforced and partially super-absorbed, showing necrosis and hemorrhaging. The tumor borders the right middle lobe (arrows) and pericardium (arrowheads), suggesting an invasion. The pleural effusion was 40 Hounsfield units, suggesting a hemorrhage within the tumor.

hemorrhaging within the tumor. We present this case in accordance with the CARE reporting checklist (available at https://acr.amegroups.com/article/view/10.21037/acr-23-44/rc).

Case presentation

A 70-year-old woman presented 2 days prior persistent right chest and shoulder pain. She had a history of hypertension. The respiratory status was stable. Enhanced computed tomography (CT) of the chest revealed the presence of a mass occupying the anterior mediastinum and a right pleural effusion. The interior of the tumor was unenhanced and partially hyper-absorbed, exhibiting necrosis and hemorrhaging. The pleural effusion Hounsfield unit (HU) was 40, suggesting hemorrhaging within the tumor (*Figure 1*). Hematological examinations revealed neutrophilic leukocytosis and elevated C-reactive protein levels. The carbohydrate antigen-125 (CA-125) and cytokeratin 19 fragment (CYFRA) levels were elevated, and the alpha-fetoprotein (AFP), human chronic gonadotropin (hCG), and anti-acetylcholine receptor antibody levels were within the normal range. Judging from the frequency at that time, the mass was diagnosed as an anterior mediastinum tumor, in particular, a ruptured/bleeding teratoma. The elevated levels of inflammatory markers and the presence of tumor necrosis on CT were suggestive of infection. Therefore, as the general status of the patient was stable and she initially received antibiotic therapy, an improvement in the inflammatory marker levels was observed with antibiotic therapy.

A surgical resection was performed 10 days after admission. The patient was placed in a dorsal position under general anesthesia. Median sternotomy revealed a tumor extending from the mediastinum to the right thoracic cavity. A bloody pleural effusion was confirmed. Since the adhesion was strong and tumor invasion was suspected, the tumor was completely resected by combining a partial resection of the right middle and lower lobes with the pericardium. Although the pleural fluid was not collected intraoperatively for cytological examinations, we judged that a gross R0 resection was achieved.

No complications occurred postoperatively and the patient was discharged from the hospital on postoperative day 14. The patient was followed up without any adjuvant therapy and has remained recurrence-free for 1 year postoperatively.

Grossly, the tumor was a 65 mm × 60 mm × 30 mm mass that contained blood clot and necrotic material with extensive adhesion to the resected lung. Microscopically, it comprised a thymoma and a thymic cyst. Hematoxylin-eosin staining revealed thick cyst walls with fibrosis, necrosis, hemosiderosis, and hemorrhaging. These findings suggested that recurrent hemorrhaging and inflammation had occurred within the tumor.

The solid components of the tumor comprised epithelial

Highlight box

Key findings

- This report describes a case wherein recurrent hemorrhage of the tumor resulted in the rupture of a thymoma.

What is known and what is new?

- Mediastinal tumors are usually discovered incidentally on chest radiography. However, rupture of a thymoma is rare.
- The pathological findings revealed the mechanism of the rupture of the tumor. An increase in the intratumoral pressure due to recurrent hemorrhaging and weakening and thinning of the tumor wall due to chronic inflammation and necrosis may have resulted in the rupture.

What is the implication, and what should change now?

- The intratumoral pressure increased due to recurrent and repeated hemorrhaging within the tumor. In addition, chronic inflammation and necrosis weakened the tumor wall. These changes contributed to the subsequent rupture of the thymoma.

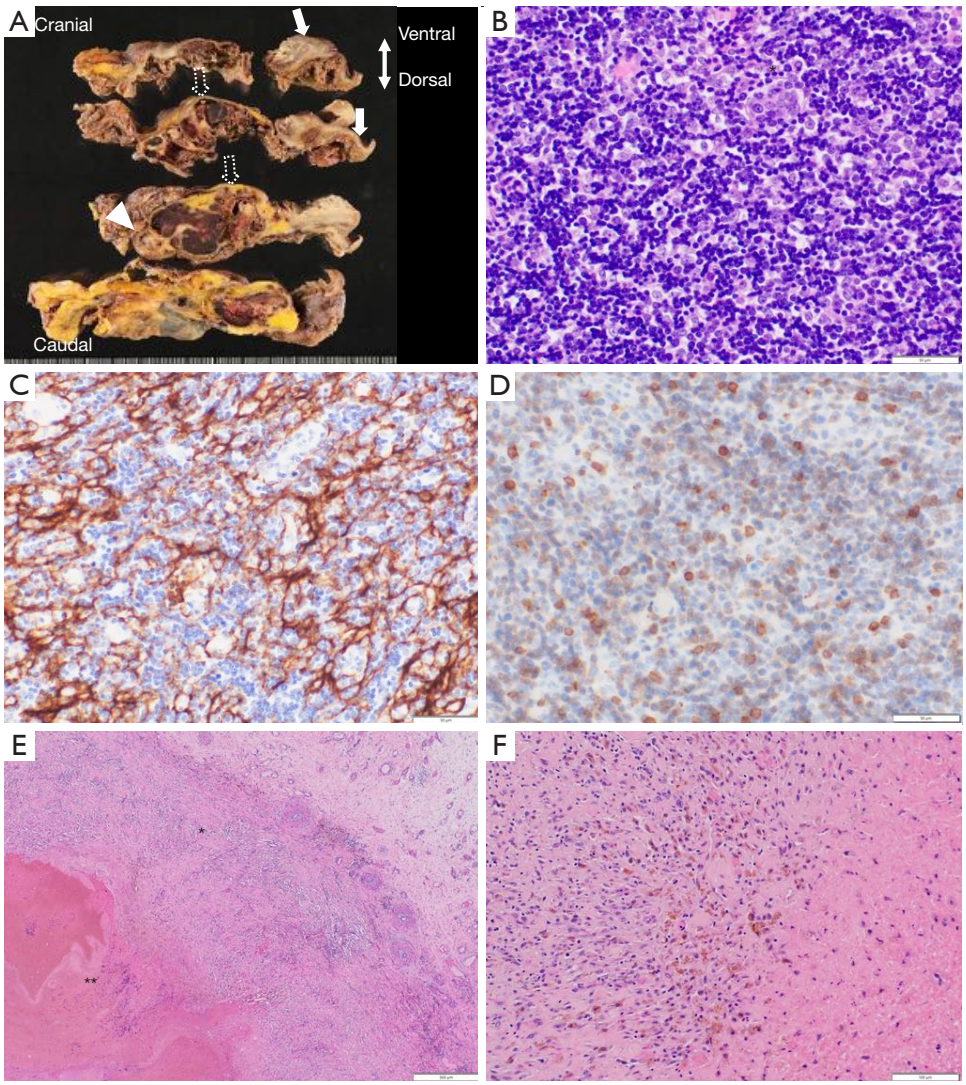

Figure 2 Tumor pathology. The cut surfaces of a formalin-fixed specimen. The right lung was partially resected (arrows). Extensive hemorrhaging and fibrosis constituted the tumor (arrow dots). The tumor contained small amounts of solid components (arrowheads) (A). Microscopically, hematoxylin-eosin staining showed that epithelial cells and lymphocytes made up the tumor. The epithelial cells formed clusters (asterisk), suggesting a type B2 thymoma (B: ×400). Epithelial cells were reticularly highlighted with cytokeratin AE1-AE3 immunostaining (C: ×400), and the lymphocytes were highlighted with leukocyte common antigen immunostaining (D: ×400). The thymic cyst wall was thick with fibrosis (*) and hemorrhaging (**) with hematoxylin-eosin staining (E: ×40). A hemosiderosis was observed in the granulation tissue, indicating that bleeding and inflammation had repeatedly occurred within the tumor with hematoxylin-eosin staining (F: ×200).

cells that had proliferated mixed with lymphocytes. The epithelial component had a highlighted mesh-pattern by cytokeratin AE1-AE3 immunostaining. These findings were suggestive of a type B2 thymoma. No tumor cells were observed in the resected lung or pericardium. The pathological diagnosis was a type B2 thymoma (World Health Organization 2020 classification), Masaoka Stage II, pT2N0M0 Stage II (Union for International Cancer Control eighth edition) (*Figure 2*).

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case

report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Mediastinal tumors are usually asymptomatic tumors that are discovered incidentally on chest radiographs (2). Rupture of teratoma occurs rarely (3). A rupture of a thymoma is extremely rare. According to previous case reports, most patients with ruptures thymoma experience sudden chest pain or dyspnea (4,5), accompanied by bleeding from the tumor and demonstrated a hemothorax. That was induced and required an emergency surgical resection (6,7).

In the present case, the patient presented with sudden-onset of right chest and shoulder pain. The high HU on CT and intraoperative right bloody pleural effusion suggested bleeding and rupture of the tumor. Since infection of the tumor was suspected, antibiotic therapy was initiated first. An elective surgery instead of emergent surgery was performed as the general status of the patient was stable.

Chest radiography revealed that the right pleural effusion had not increased over time, indicating that the hemorrhaging was temporary and hemostasis had been achieved. The pathological findings revealed the mechanism of the rupture of the tumor. Hemorrhaging, fibrosis, necrosis and hemosiderosis were observed within the tumor.

Based on these findings, we hypothesized that the following factors may have contributed to the rupture. First, recurrent hemorrhaging and inflammation had occurred within the tumor, resulting in fibrosis and hemosiderosis during the chronic phase. Consequently, the intratumoral pressure may have increased, which could have been the main cause of the thymoma rupture. Second, chronic inflammation and necrosis may have led to weakening of the surrounding wall and cyst wall, resulting in the rupture of the thymoma. In present case, part of the tumor consisted of the cyst wall which may easily rupture under the circumstances of hemorrhage or inflammation (8).

The reasons for the rupture of thymoma remain obscure due to its rarity.

According to the previous studies, an enlargement of the thymoma (6) and intratumoral hemorrhaging (9) were possible factors for the rupture of thymoma. In the present case, recurrent hemorrhaging and inflammation and necrosis were considered to have contributed to the rupture of the thymoma.

Our case report presents the mechanism of the rupture of thymoma from pathological findings.

Conclusions

We here reported a case of ruptured thymoma. Based on the findings of our case, it can be considered that the recurrent and repeated hemorrhaging of the tumor may have increased the intratumoral pressure and chronic inflammation and necrosis weakened tumor wall. These changes contributed to the subsequent rupture.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://acr.amegroups.com/article/view/10.21037/acr-23-44/rc

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://acr.amegroups.com/article/view/10.21037/acr-23-44/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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