In conclusion, endotracheal Castleman’s disease is a very rare disease that can cause respiratory distress. Rigid bronchoscopy with APC can be an effective and safe technique for treating endotracheal CD.

References

Spontaneous Thymic Hemorrhage in an Adult
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Spontaneous thymic hemorrhage in a normal thymus in neonates and infants has been reported in the

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1800 CASE REPORT SAKURABA ET AL. SPONTANEOUS THYMIC HEMORRHAGE

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literature. Only one case of spontaneous thymic hemorrhage in an adult has been reported to our knowledge. We herein report the case of an adult who had a cardiac operation 26 years previously and who was on anticoagulation. He experienced acute hemorrhage in a normal thymus, and this was not thought to be attributable to an accidental cause such as trauma or to hypertension.


Spontaneous thymic hemorrhage in a normal thymus in an adult is very rare, with only 1 case report to our knowledge [1]. We herein report an adult who was found to have spontaneous thymic hemorrhage in a normal thymus, who had 26 years previously had cardiac surgery and was on anticoagulation.

To our knowledge, this case is the first report of spontaneous thymic hemorrhage in an adult after cardiac surgery.

The patient was a 44-year-old man who had an aortic valve replacement 26 years earlier. He had since been on anticoagulation to maintain an international normalized ratio of 2.5. He was known to be normotensive. Chest computed tomography was normal 6 years before his presentation at our institution (Fig 1). He experienced sudden-onset left-side chest pain and went to the hospital. He reported no history of trauma. Chest roentgenograph revealed a large shadow in the anteroposterior window (Fig 2). Chest computed tomography revealed multiple soft tissue masses in the anterior mediastinum (Fig 3A). Suspecting thymic hemorrhage, his anticoagulation was stopped. The patient was monitored in the hospital, and serial enhanced chest computed tomography showed that the mass was progressively enlarging 18 days after his initial imaging on presentation (Fig 3B). We proceeded with thoracotomy, which demonstrated an enlarged hemorrhagic thymus. A total thymectomy was performed. Pathologic examination revealed a normal thymus with significant hemorrhage (Fig 4A). There was also hemorrhage in the medulla, with scattered thymic tissue (Fig 4B). The postoperative course was uneventful.

**Comment**

Causes of hemorrhage in the mediastinum are numerous, with case reports of bleeding owing to thymic neoplasm, uremia, and parathyroid adenomas, and occurring after cardiac catheterization and in hemophiliacs. Wooly and colleagues [2] reported 2 cases in infants, Siger and coworkers [3] reported 1 case in an infant, Bees and associates [4] reported 1 case in a neonate, and Sakusenberg and colleagues [5] reported a case of an intrauterine fetal death. All of these patients had defects in coagulation.

Fisher and Reis [6] reported the case of an 8-year-old girl who exhibited cardiac tamponade owing to acute thymic hemorrhage shortly after open heart surgery for the correction of congenital cardiac anomalies. In this case, the cause of bleeding was after surgical complications. In our case, cardiac surgery was performed 26 years before the thymic hemorrhage; the cause of bleeding was therefore unrelated to the surgery.

Ghoshhajra [1] reported a spontaneous thymic hemorrhage in an adult in 1977, and that is the first report in an adult to our knowledge. The patient had no defects in coagulation. Our case is the first report to our knowledge of spontaneous thymic hemorrhage in an adult after cardiac surgery.

Our patient had a history of a cardiac operation 26 years previously and had since been therapeutically...
anticoagulated, a known cause of spontaneous thymic hemorrhage. However, there are many patients worldwide who undergo cardiac operations with subsequent anticoagulation, with few or no reports of spontaneous thymic hemorrhage. The risk associated between this phenomenon and anticoagulation is therefore not known.

Alternatively, patients with spontaneous thymic hemorrhage could have thymic disease predisposing them to bleeding, such as a thymic cyst. In our case, however, chest computed tomography was performed 6 years previously and was normal. Microscopically, a cystic wall did exist in conjunction with the hemorrhage. However, in neighboring normal thymic tissue hemorrhage was also seen, particularly in the medulla. For this reason, we conclude that this was a case of idiopathic spontaneous thymic hemorrhage in a normal thymus.

References

Recurrent Thymoma With Stiff-Person Syndrome and Pure Red Blood Cell Aplasia
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Stiff-person syndrome (formerly known as stiff-man syndrome) is a very rare autoimmune and neurogenic disorder, thought to present as a paraneoplastic variant in association with thymoma. Pure red blood cell aplasia is also a paraneoplastic disorder associated with thymoma. Although separate cases of stiff-person syndrome and pure red blood cell aplasia have been reported, we describe here what is to our knowledge the first case of recurrent thymoma with both stiff-person syndrome and pure red blood cell aplasia. We describe the successful treatment of the neurogenic symptoms of stiff-person syndrome and the progressive anemia associated with pure red blood cell aplasia by tumor excision.

Fig 3. Chest computed tomography. (A) Initial presentation. There is a soft tissue mass in the anterior mediastinum. (B) Eighteen days later, the mass is progressively larger, and the area surrounding the mass is enhanced.

Fig 4. Pathologic findings. (A) Resected specimen shows hemorrhagic cavity in the thymus. (B) Microscopic findings shows hemorrhage amid the medulla of thymic tissue (hematoxylin-eosin stain, magnification ×40).

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