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Sudden death in a child caused by a giant cavernous hemangioma of the anterior mediastinum

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ABSTRACT

A 4-year-old girl who had been treated for asthma since the age of 2 years had a severe coughing fit and died suddenly. The patient had a history of occasional severe coughing fits, and these fits had been worsening in severity during the week before her death. Prior to death, she was taken to a clinician, and thymic hypertrophy was suspected based on chest X-ray findings. The clinician recommended that she visit a general hospital at a later date; however, she died that night. Postmortem radiology and autopsy revealed a large mass in the anterior mediastinum compressing the heart and airway, and no other findings attributable to sudden death were observed. Therefore, we concluded that the patient's death was attributable to acute respiratory and cardiac circulatory failure secondary to the pressure induced by the mass. Microscopically, the mass showed a cavernous structure composed of cystically dilated, thin-walled large vessels filled with blood. The final diagnosis was a cavernous hemangioma. Hemangiomas are the most common benign vascular anomalies seen in young children; however, mediastinal hemangiomas are rare and can cause life-threatening complications because of their size and location. Therefore, forensic pathologists should include hemangioma as a differential diagnosis in children with anterior mediastinal masses.

1. Introduction

Hemangiomas are the most common vascular anomalies in infants, and they occur in about 10% of term infants.[1] More than half of them are located in the head and neck region,[1] and mediastinal hemangiomas are rare, with an incidence of <0.5% of all mediastinal tumors.[2] Most hemangiomas do not require specific treatment,[1] because they are benign and almost never become malignant. However, they can become very large and even fatal if they affect vital structures.[1,3] Microscopically, hemangiomas are divided into two broad categories: capillary hemangiomas and cavernous hemangiomas.[4] In contrast to capillary hemangiomas, cavernous hemangiomas do not regress spontaneously,[3] and the progress of the tumor should be carefully observed.[5]

We herein report a fatal case of a giant cavernous hemangioma of the anterior mediastinum in a child, in which the mass severely compressed the heart and airway. The patient presented with worsening coughing and died suddenly without a clinical diagnosis; therefore, an autopsy was required. We believe that this case is worth reporting because few cases of sudden death caused by mediastinal hemangiomas have been reported in literature.

2. Case report

A 4-year-old girl was taken to a private clinician because of a 1-week worsening history of coughing. The patient had a history of intermittent high fever and severe coughing fits since the age of 2 years, and she had been diagnosed with asthma. She had been seen in the clinic every few weeks, and treated with inhaled asthma medications. On that day, thymic hypertrophy was suspected based on chest X-ray findings, and the clinician recommended that she visit a general hospital at a later date. She looked well when she went home, and ate up dinner. She went to bed with her father, and 1 h later, she began coughing severely and developed dyspnea. Her father rubbed her back, however, she stopped breathing immediately. He started cardiopulmonary resuscitation and called an

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ambulance. She was in cardiopulmonary arrest when the ambulance crews arrived. She was taken to the emergency room and underwent resuscitation procedure for about an hour, however, could not be resuscitated. The time interval between the onset of cardiopulmonary arrest following severe coughing at home and declaration of death in the ER was approximately 1.5 h. She did not have return of spontaneous circulation during this entire time period.

The patient had no significant medical illness except asthma, and an autopsy was performed for determining the cause of death.

3. Postmortem radiology

As a part of the pre-autopsy screening, postmortem radiographs were obtained with a mobile plain X-ray system (MobileDaRt Evolution; Shimadzu Corporation, Kyoto, Japan) integrated with a digital flat-panel detector (CXDI-50G; Canon Inc. Medical Equipment Group, Tokyo, Japan) approximately 13 h after the confirmation of her death. We also used a 64-channel multidetector computed tomography (CT) machine (Aquilion; Toshiba Medical Systems, Tokyo, Japan) and an open permanent magnet-type magnetic resonance imaging (MRI) scanner (AIRIS Vento; Hitachi Medical Corporation, Tokyo, Japan).

The chest X-ray revealed that the right lower lobe and left upper lobe maintained a small volume of air, and almost all lung areas showed opacification (Fig. 1). The cardiac and mediastinal contours and the radiolucent structure of the gas-filled trachea were not identified (Fig. 1). On postmortem CT and MRI, a giant mass occupied the anterior mediastinum, compressing the heart toward the left dorsal side and deforming the tracheal rings (Fig. 2). The mass exhibited a multilocular cystic structure with a fluid–fluid level, suggesting that it contained blood (Fig. 2). No lethal intracranial hemorrhage or abdominal injury was present. No upper- or lower-extremity fractures were confirmed.

4. Autopsy findings

The patient's body was 106 cm in height and 14 kg in weight. Other than some needle marks on her extremities, there were no apparent antemortem injuries. The face was not congested, and there were no petechial hemorrhages in the palpebral conjunctivae.

A 13- × 13- × 7-cm mass was present in the anterior mediastinum, partly adjacent to the thymus (Fig. 3). The thymus was shrunken and weighed 13 g. The mass contained a large volume of bloody fluid, and the cut surface showed a multilocular cystic appearance (Fig. 4).

The heart weighed 100 g, and there were no congenital abnormalities. The lungs (both 130 g) were moderately congested and edematous. Lung hyperinflation and mucous plugs were not seen. There was a small amount of pale red mucous in the airway. The luminal surface of the trachea and the main bronchus were intact. No abnormal findings were apparent in the other organs.

5. Histopathology

The mass showed a cavernous structure composed of cystically dilated, thin-walled large vessels filled with blood (Fig. 5). The thymus was histologically normal. Most of the pulmonary parenchyma was collapsed; however, overexpansion of the alveoli was seen in a small area. Mild thickening of the bronchial basement membrane was seen, and a few eosinophils were present in the mucosa. No bronchial mucus plugs, goblet cell hyperplasia, hyperplasia of mucosal and submucosal vessels, mucosal edema, or bronchial smooth muscle hypertrophy were seen.

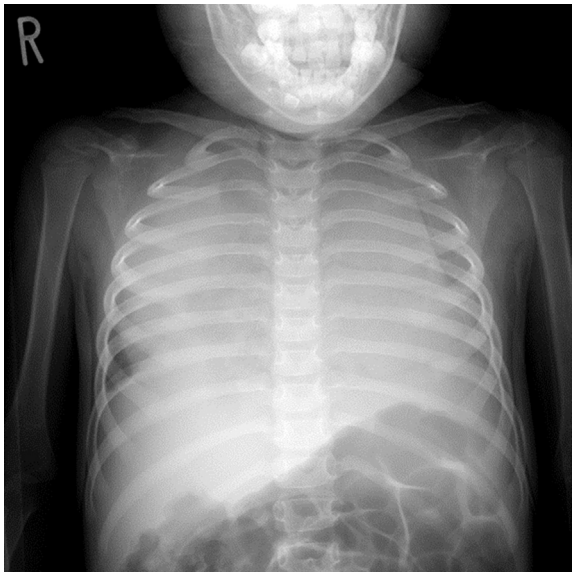

Fig. 1. Postmortem anteroposterior chest X-ray.
Both lungs exhibited areas of decreased lucency. The cardiac shadow and radiolucent structure of the trachea were not found.

6. Discussion

Postmortem radiology and autopsy revealed a large mass in the anterior mediastinum, and the mass severely compressed the heart and airway. The heart was compressed toward the left dorsal side, and the tracheal rings were distorted (Figs. 1 and 2).

The mass showed a cavernous structure composed of cystically dilated, thin-walled large vessels filled with blood (Fig. 5), therefore it was diagnosed as cavernous hemangioma. Hemangiomas are the most common vascular anomalies of infancy, and they occur in about 10% of term infants.[1] More than half of them are located in the head and neck region[1]; mediastinal hemangiomas are rare, with an incidence of <0.5% of all mediastinal tumors.[2] Most hemangiomas do not require specific treatment; however, approximately 10% of them are symptomatic, and 1% are life-threatening because their location around the airway can cause airway obstruction.[1]

Almost 50% of mediastinal hemangiomas are asymptomatic.[6] Symptomatic patients present with coughing, chest pain, fever/chills, and dyspnea.[5–8] The symptoms are correlated with age, and younger patients tend to have more severe symptoms.[7] These symptoms are generally caused by direct invasion to adjacent structures[6]; however, the tumor in the present case was not invasive, and the internal surface of the trachea was intact. Therefore, the symptoms in our case seemed to be caused by the pressure induced by the mass, not by direct invasion.

[7] Hemangiomas are classified as follows according to their clinical appearance and the caliber of vessel involved: capillary hemangiomas, cavernous hemangiomas, large vessel hemangiomas, skeletal muscle hemangiomas, intravascular papillary endothelial hyperplasia, spindle cell hemangiomas, and hobnail