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Case Report

Intramyocardial bronchogenic cyst: histological appearance and a review of the literature

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ABSTRACT

We report histological appearance of a bronchogenic cyst that was incidentally found in a 78-year-old man who died from drowning related to a traumatic accident. The cyst was found in the posterior edge of the interventricular septum and was monolocular with a 5-mm diameter. The cystic wall was not associated with cartilage or the smooth muscle layer, and was lined by ciliated respiratory epithelium. Immunohistochemistry showed that many of these epithelial cells were positive for cytokeratin 7 and thyroid transcription factor-1. A few of the cells were positive for CA19–9, chromogranin A, synaptophysin, and S-100. No cells were positive for cytokeratin 20, p63, bcl-2 and napsin A. This feature was compatible with bronchogenic cyst, and is not common compared with previously reported immunohistochemical features of cystic tumors of the atrioventricular node. A low prevalence of p53, single strand-DNA, and Ki-67 indicated modest cell turnover in the epithelial cells of present case. Our survey of the English literature showed 23 cases of intramyocardial bronchogenic cyst. Although asymptomatic cases were dominant in the literature, some intramyocardial bronchogenic cysts showed life-threatening arrhythmia, which could cause sudden unexpected death.

1. Introduction

Bronchogenic cyst is a congenital malformation, which arises from abnormal budding of the ventral diverticulum of the embryonic foregut. Besides the mediastinum, intrapulmonary region, neck, diaphragm, intraabdominal region, and intramedullary part of the spine, bronchogenic cyst is also rarely found in the heart, depending on the embryonic stage of development at which the anomaly occurs [1–3]. In the heart, many patients show such cysts in the pericardium and they are rarely observed in the intramyocardial region [4]. The cardiac primordial is located in close proximity to the foregut, and abnormal budding of the foregut may rarely migrate to the myocardium as a source of this intramyocardial cyst [1]. We report the histological appearance, including immunohistochemistry, of a case of intramyocardial bronchogenic cyst. We compared our case with previously reported immunohistochemical appearances of a cystic tumor of the atrioventricular node (CTAVN). A CTAVN is a benign cystic mass of unknown etiology and is associated with atrioventricular block in some cases. We also summarize previously reported cases in English to promote better understanding of this rare pathological condition.

2. Case description

A 78-year-old man was found dead in a car that had driven into a river. According to a witness, the car suddenly swerved off a road and subsequently drove into the river without slowing down. On admission, he was found to have suffered from cardiopulmonary arrest, and resuscitation was not successful. An autopsy showed clear signs of drowning, such as marked edematous swelling of both lungs and plenty of whitish froth in the airways, and mild bruising of the chest by possible contusion from the steering wheel. The blood-ethanol concentration was 0.7 mg/ml. An electrocardiogram examined 1 year before his death showed transient atrial fibrillation and a small number of premature ventricular complexes. Transthoracic echocardiography did not show any major abnormal findings. He was continuously medicated by β-blockers and anticoagulants.

The heart weighed 450 g. We found mild left ventricular hypertrophy, atherosclerosis of the coronary artery without significant stenosis, and mitral ring calcification. The heart was fixed in 10% buffered formalin. A unilocular cystic lesion was found in the base of the interventricular septum in the process of dissecting the specimen containing the atrioventricular conduction system. The cyst was 5 mm in diameter and was filled by whitish mucinous fluid (Fig. 1a and b). The cystic cavity in the interventricular septum was lined by ciliated columnar epithelium with pseudostratification in part of the epithelium, on

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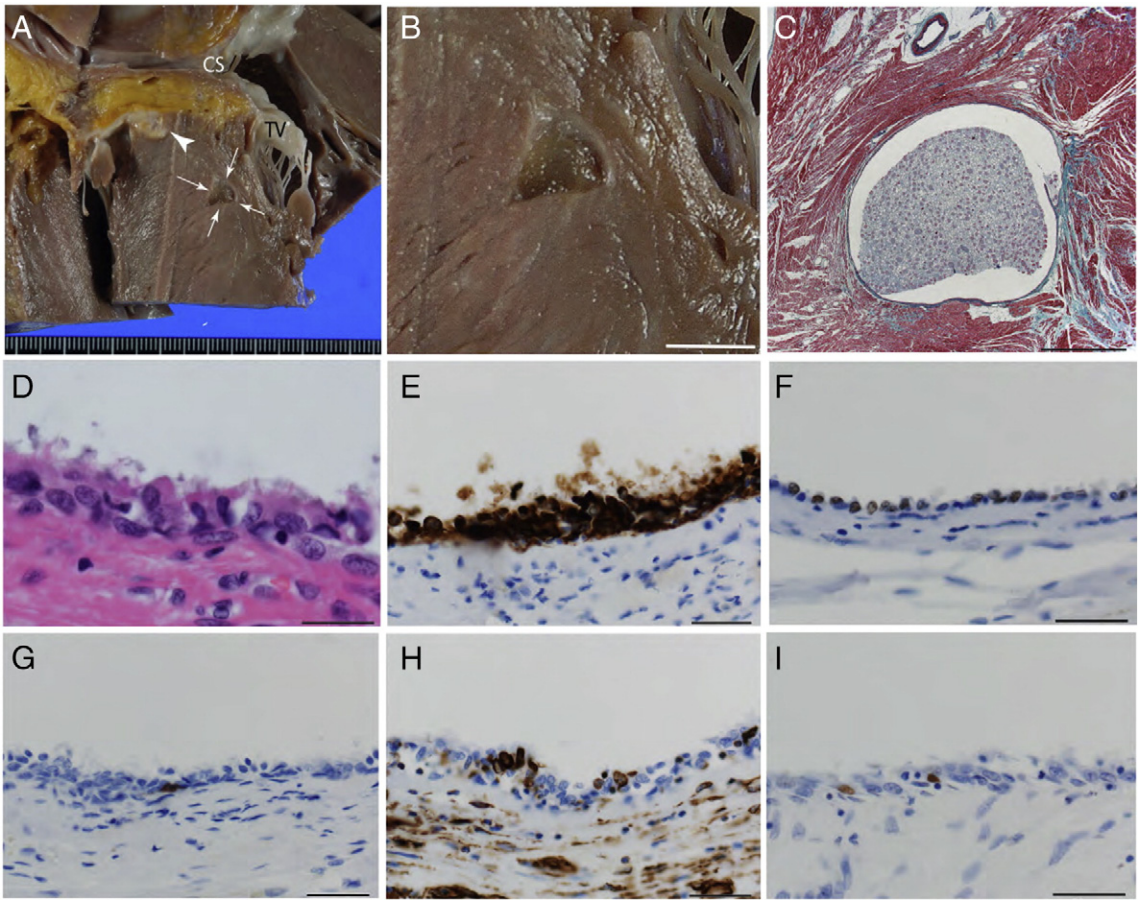

Fig. 1. Pathological findings of the bronchogenic cyst. (A) Gross appearance of a longitudinal section of the posterior basilar ventricular septum. A unilocular cystic lesion in the interventricular septum is indicated by four arrows. The arrowhead indicates mitral ring calcification. TV, tricuspid valve; CS, coronary sinus. (B) Image of intramyocardial bronchogenic cyst containing gelatinous material. Scale bar=5 mm. (C) Low-power image of intramyocardial bronchogenic cyst. Scale bar=2 mm. (D) High-power image of lining epithelium. Cuboid respiratory epithelium with cilia can be seen. Scale bar=10 μm. (E–I) Immunohistochemistry. Lining epithelium shows a lot of positive staining for CK7 (E) and TTF-1 (F), and sparsely positive staining for chromogranin A (G), S-100 (H), and ss-DNA (I). Scale bar=20 μm.

microscopically. Smooth muscle and cartilage were not evident in the cystic wall. Some of the cells also showed apocrine secretion. Mild circumferential fibrosis was found, but no inflammatory focus was evident (Fig. 1c and d).

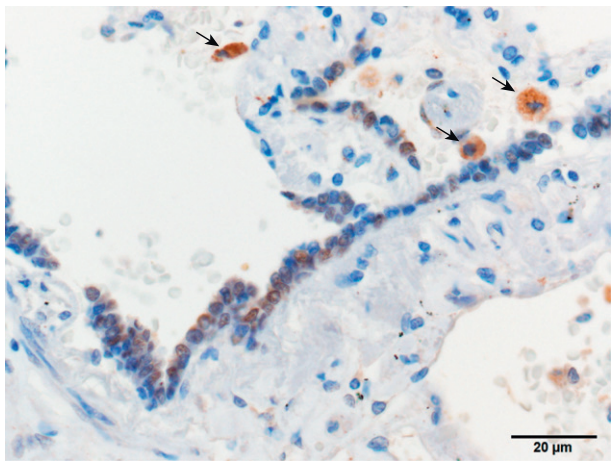

Fig. 2. Immunohistochemistry using a cocktail of antibodies, including anti TTF-1 and napsin A, in the control case. TTF-1 is positive for the nucleus of bronchiolar epithelium and napsin A is positive for the cytoplasm of alveolar macrophages (arrows). Scale bar=20 μm.

Immunohistochemical staining was performed for cytokeratin (CK) 7 (OV-TL 12/30; 1:200; DAKO, Glostup, Denmark), CK 20 (Ks20.8; 1:100, DAKO), CK AE1/AE3 (AE1/AE3; 1:200, DAKO), CK high molecular weight (CK-HMW) (34βE12; 1:100, DAKO), CK-CAM5.2 (CAM5.2; 1:16, Becton Dickinson, Franklin Lakes, NJ), Vimentin (V9; 1:800, DAKO), carcinoembryonic antigen (CEA) (11–7; 1:100, DAKO), CA19–9 (1116-NS-19-9; 1:200, DAKO), chromogranin A (LK2H10; predilute X2, Roche mtm Laboratories AG, Manheim, Germany), synaptophysin (27G12; predilute X2, NICHIREI, Tokyo, Japan), S-100 (S-100, Leica Biosystems, Bannockburn, IL), α-Smooth muscle actin (α-SMA) (1A4; predilute X4, DAKO), Calretinin (PAD:DC8; predilute X4, Zymed Laboratories Incorporated, San Francisco, CA), WT1 (6F–H2; predilute X 2, Roche), p63 (4A4; predilute X2, NICHIREI), bcl-2 (124; 1:200, DAKO), Thyroglobrin (DAK-tg7; 1:10,000, DAKO), estrogen receptor (ER, 1D5; 1:100, DAKO), progesterone receptor (PgR, PgR636; 1:100, DAKO), p53 (DO-7; 1:100, Leica), single strand DNA (ss-DNA) (4B013A; 1:100, DAKO), ki-67 (MIB-1; 1:200, DAKO), and ADC Cocktail Antibody [TTF-1 (SPT24) and Napsin-A (TMU-Ad02); predilute X4, Pathology Institute Corp., Toyama, Japan]. Immunostaining was performed using the Leica Bond-IV automation and Leica Refine detection kit (Leica). A lung specimen that was obtained from an age-matched case (78 year-old man who died from a traffic accident) was also examined in the same manner for comparison with the present case. Epithelial cells that lined the proximal bronchus with cartilage and smooth muscle and epithelial cells that lined the distal bronchus and bronchioles were evaluated.