

Mediastinal mature teratoma with complete gastrointestinal and bronchial walls

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Keywords

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Abstract

An extremely rare case of anterior mediastinal mature teratoma with almost complete gastrointestinal and bronchial walls is described. A 65-year-old woman presented with left precordial pain. Chest computed tomography showed a huge anterior mediastinal tumor, 15 cm × 21 cm, occupying the left thoracic cavity. Post-resection histopathological examination confirmed the diagnosis of mature teratoma and demonstrated almost complete gastrointestinal and bronchial walls. Although mature teratomas of the ovary and sacrococcygeal area are known to rarely contain organoid structures with various degrees of differentiation, this is the first case of an anterior mediastinal mature teratoma that contained well-developed organoid structures.

Introduction

Mature teratomas are composed of several foci of mature tissues derived from at least two of three embryonic germ layers. Mature tissues derived from each type of embryonic germ layer usually have no correlation with the others [1], such that gastrointestinal mucosa or bronchial mucosa derived from endoderm and smooth muscle tissues derived from mesoderm are contained independently in the majority of mature teratomas [2]. Only a few cases of mature teratoma arising from the ovary or sacrococcygeal area describing well-developed organoid structures including gastrointestinal walls have been reported previously [3, 4]. The first report of a case of a mature teratoma of the anterior mediastinum in which mucosa coordinated with smooth muscle which resulted in the organizing of almost complete gastrointestinal and bronchial walls forming multilocular cysts is reported.

Case Report

A 65-year-old woman without a significant medical history presented to her regular doctor due to left precordial pain. A chest radiograph showed a huge mass in her left hemithorax with a contralateral shift of mediastinal structures and she was thus referred to our hospital. On chest computed tomography (CT), a huge anterior mediastinal tumor that measured 15 cm × 21 cm mostly occupying the left thoracic cavity was seen. The tumor contained multilocular cysts that were characterized by slightly increased attenuation on contrast-enhanced examination (Fig. 1A). Calcification and fat attenuation were noted in the solid areas, which suggested the diagnosis of mature teratoma. Its cyst walls were rather strongly visualized on enhanced magnetic resonance imaging (MRI) (Fig. 1B). Because the clinical presentation of the patient indicated the possibility of impending rupture, surgery was performed.

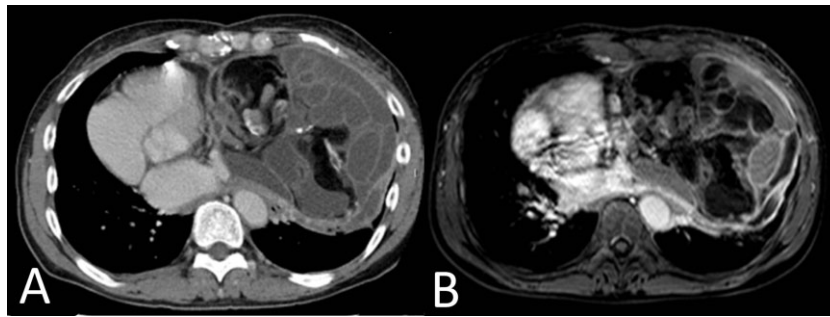


Figure 1. (A) Chest computed tomography shows that the tumor contains slightly enhanced multilocular cysts. (B) Enhanced magnetic resonance imaging facilitates the visualization of the cyst walls.

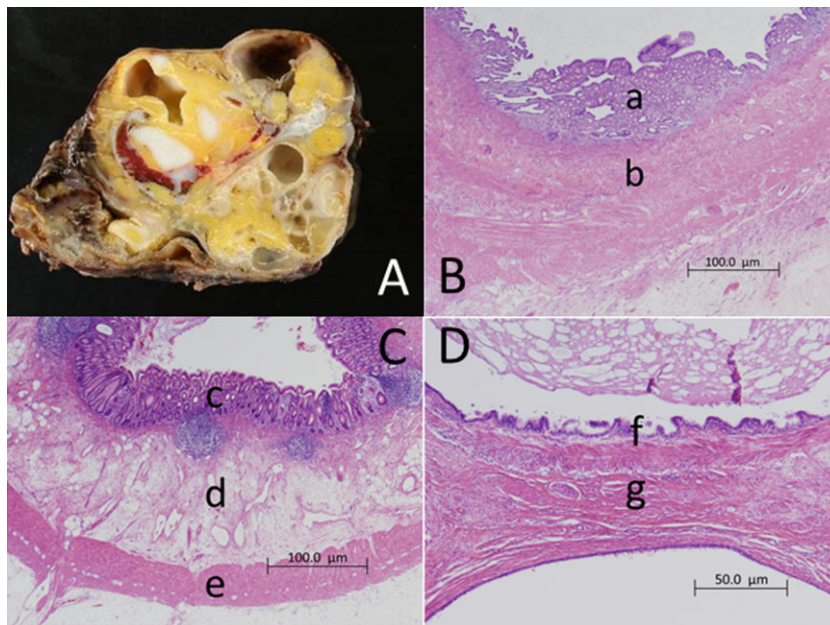


Figure 2. (A) The gross appearance of the specimen is associated with multilocular cystic lesions. (B) The stomach wall cysts demonstrate gastric mucosa with a fundic gland (a) and two layers of muscularis propria (b) (hematoxylin and eosin staining, original magnification $\times 20$). (C) The colonic wall cyst consists of mucosa with intestinal crypts (c), submucosa with lymph follicles (d), and two layers of muscularis propria including Auerbach's plexus (e) (hematoxylin and eosin staining, original magnification $\times 20$). (D) The bronchus wall cysts have ciliated epithelium (f) and a muscular layer (g) (hematoxylin and eosin staining, original magnification $\times 40$).

The operation was started with a median sternotomy, but the huge size of the tumor required suction of the fluid component of the cystic regions and an additional intercostal thoracotomy at the left second intercostal space to ensure sufficient working space. The tumor showed dense adhesions to the lingular segment of the left upper lobe and the pericardium, which resulted in the need to resect the tumor with these structures.

The cut surface of the specimen showed multilocular cystic areas filled with mucinous fluid (Fig. 2A). Histopathological examination demonstrated the multilocular cysts that corresponded to almost complete gastrointestinal and bronchial

walls: stomach walls, colonic walls, and bronchus walls. The stomach wall cysts included gastric mucosa with fundic glands and two layers (circumferential and longitudinal) of muscularis propria (Fig 2B); similarly, the colonic wall cysts included mucosa with intestinal crypts, submucosa with lymph follicles, and two layers of muscularis propria with Auerbach's plexus (Fig. 2C). Thus, the stomach wall cysts and colonic wall cysts reproduced almost the complete walls of their original organs, but without the serosa. The bronchus wall cysts also possessed ciliated epithelium combined with a muscular layer (Fig. 2D). In the solid areas, bone tissues, nerve tissues, and skin with its appendages were observed,

confirming the diagnosis of mature teratoma. No malignant or immature component was found.

Discussion

An extremely rare case of mature teratoma of the anterior mediastinum associated with almost complete gastrointestinal and bronchial cysts was described.

Mediastinal mature teratomas are benign neoplasms that usually arise in or near the thymus and account for 70–75% of primary germ cell tumors of the mediastinum [2, 3]. In adult females, all germ cell tumors of the mediastinum are mature teratomas in principle and they are mostly cystic [2]. The cystic regions are lined with keratinized or glandular epithelium in usual.

In the current case, the characteristic finding of multilocular cysts with increased visualization on contrast-enhanced CT and MRI that corresponded histopathologically to almost complete gastrointestinal and bronchial cysts was demonstrated.

As a differential diagnosis of cystic regions with organized gastrointestinal and bronchial walls in the anterior mediastinum, bronchopulmonary foregut malformation (BPFM) was considered.

The concept of BPFM is defined to include a wide spectrum of congenital foregut malformations. A supernumerary lung bud arising from the foregut during the fourth to fifth week of embryogenesis is considered the etiology, and therefore, the cystic structures observed in BPFM are composed of organized gastrointestinal and bronchial walls without ectodermal components [5]. However, in the present case, the tumor contained skin and nerve tissues, both derived from ectoderm, so that the diagnosis of mature teratoma was confirmed.

On the other hand, a theory that teratomas arise from dislocated totipotent primordial germ cells (PGCs) misplaced during their migration from the yolk sac endoderm to the gonad during early embryogenesis is widely accepted [1]. That is, in mature teratomas, PGCs misplaced in an ectopic environment differentiate into mature tissues via three embryonic germ layers. Commonly, there is no interactive coordination with each germ layer component, resulting in disorganized admixtures of mature tissues [1]; therefore, the presence of organoid structures in which

mucosa coordinated with smooth muscle tissues to organize almost complete gastrointestinal and bronchial walls, as in the current case, is a rare finding. Few previous reports have shown rare cases of mature teratomas arising from the ovary or sacrococcygeal area with organoid structures having various degrees of differentiation. The presence of almost complete intestinal loops in ovarian and sacrococcygeal mature teratomas has been demonstrated previously [3, 4]. However, no similar case of teratoma arising from the anterior mediastinum has been reported previously.

The present case is, to the best of our knowledge, the first reported case in which mature teratoma arising from the anterior mediastinum showed almost complete gastrointestinal and bronchial walls.

Disclosure Statements

No conflict of interest declared.

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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