Rare Presentation of Giant Isolated Enteric Cyst in Anterior Mediastinum

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Mediastinal enteric cysts are extremely rare. In the published English literature, most such cysts are reported in the right posterior mediastinal position, often with vertebral anomalies. Just four cases have been reported for enteric cysts in atypical mediastinal locations since 1997. This report describes an additional case of an atypically located giant isolated enteric cyst in the anterior mediastinum.

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Mediastinal enteric cysts are a rare occurrence encountered mainly in neonates and infants. More than 70% of enteric cysts manifest as a right posterior mediastinal mass, often accompanied by vertebral defects [1]. The published English literature contains just four cases of atypically located enteric cysts in the mediastinum. Here we report an additional presentation of a giant isolated enteric cyst in the anterior mediastinum with ambiguous radiologic features.

A 27-year-old woman was incidentally found to have a mediastinal mass on a chest roentgenogram. She had no symptoms, and physical examination was unremarkable. A computed tomographic (CT) scan revealed the presence of a large, nonenhanced, multilocular, homogeneous

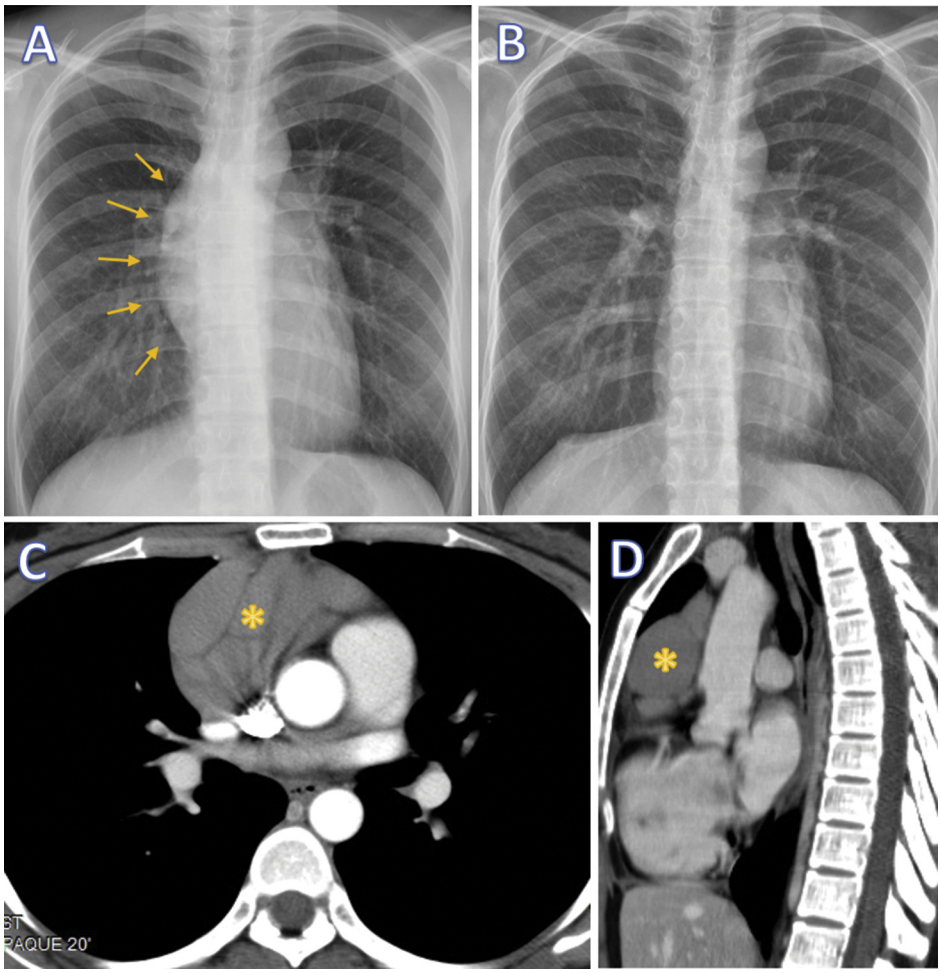

Fig 1. A 27-year-old female patient was found incidentally to have an asymptomatic mediastinal mass. The chest roentgenogram demonstrated (A, arrows) obvious mediastinal widening with a positive incomplete border sign. A computed tomography (CT) scan showed (C and D, asterisk) a large, nonenhanced, multilocular, homogeneous cystlike lesion measuring 80 × 42 × 105 mm in the anterior mediastinum. The patient underwent mediastinal mass excision by video-assisted thoracoscopic surgery and remained free from recurrence 5 years postoperatively. (A) Preoperative chest roentgenogram. (B) Postoperative chest roentgenogram. (C) Contrast-enhanced CT scan. (D) Contrast-enhanced CT scan, sagittal view.

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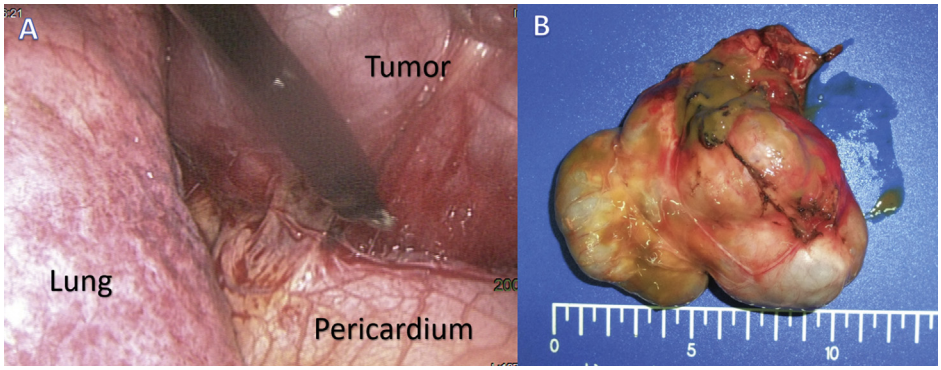

Fig 2. The tumor was excised by triportal video-assisted thoracoscopic surgery. (A) The thin-walled cyst was freely movable with only mild adhesion to the outside surface of the pericardium. (B) This cyst was filled with brown, sticky fluid and had a wall structure similar to that of mature bowel.

cystlike lesion, measuring 80 × 42 × 105 mm, in the anterior mediastinum (Fig 1). A gallium-67 scan showed no evidence of gallium-avid tumor. The patient underwent mass resection using triportal video-assisted thoracoscopic surgery. The lesion was freely movable; with only mild adhesion to the overlying surface of the pericardium (Fig 2A). The thin-walled cyst was found to be filled with brown, sticky fluid (Fig 2B). The structure and morphology of the cyst were similar to those of mature bowel. The pathology results revealed a benign cyst lining with intestinal mucosa and a muscle layer mixed with adipose tissue (Fig 3). The patient made an uneventful recovery after resection and was free from recurrence 5 years postoperatively.

Comment

Enteric cysts are generally thought to result from developmental anomalies of the embryonic primitive foregut, in particular, an incomplete separation of the notochord and primitive foregut [2]. In many cases, the affected child is subsequently born with abnormalities of the vertebral column. However, such abnormalities were not observed in the present case. Furthermore, enteric cysts are usually found in the right posterior mediastinum [1], whereas the present lesion was located in the anterior mediastinum. Enteric cyst formation may alternatively be explained by abnormal budding from the primitive foregut. In such cases, an isolated enteric cyst develops when the budding portion of the foregut detaches and migrates to another site in the body, thereby leading to enteral differentiation [2]. Our case appears to be consistent with this theory because of the conflict between the location of the lesion (the anterior mediastinum) and its suspected origin (ie, alimentary tract).

In most patients presenting with mediastinal enteric cyst, the diagnosis is made in the neonatal or infant period as a result of respiratory distress and the need for immediate surgical intervention. However, some cases are found incidentally in childhood or adulthood with an asymptomatic presentation. A review of the published English literature revealed just four cases of mediastinal enteric cyst in atypical locations (ie, other than the right posterior mediastinum). Of these four cases, two were left posterior mediastinal masses [3, 4]; one was in the middle mediastinum, mimicking a pericardial cyst; and one was in the anterior mediastinum [5, 6]. However, although Carr and colleagues [6] reported an enteric cyst in the same position as in our case (ie, anterior mediastinum) in 1977, CT technology was unavailable at that time, and hence diagnosis was not possible. In our case, the CT image revealed a structure and morphology similar to that of the intestine. Thus, enteric cyst must be taken into consideration in the differential diagnosis of anterior mediastinal tumors.

Obtaining a preoperative diagnosis of mediastinal enteric cyst is challenging. In the present case, CT-guided biopsy was unable to provide a definitive diagnosis of cystic teratoma or lymphoma. Surgical resection was nonetheless performed because of the ambiguity of the biopsy results. Some adhesion was noted between the cystic wall and the adjacent tissue, including the lung surface and pericardium. However,

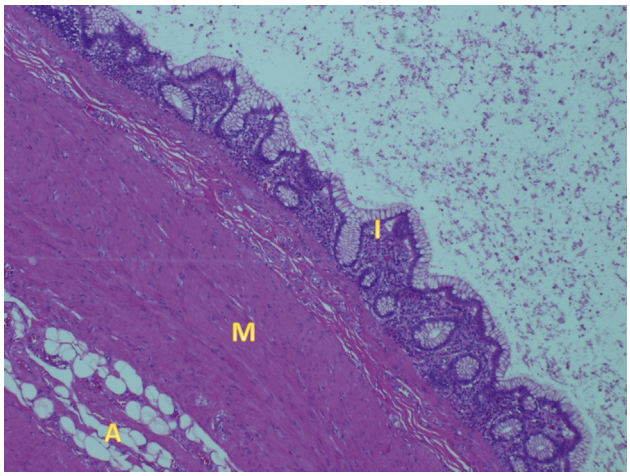

Fig 3. High-magnification view of the cyst wall. The cyst was lined with intestinal mucosa (I) and a muscle layer (M) mixed with adipose tissue (A). (Hematoxylin and eosin stain; original magnification ×100.)

the patient underwent complete resection with ease. The histologic results revealed that the structure of the cystic wall was very similar to that of mature bowel wall. Moreover, no evidence of gastric or pancreatic differentiation or malignancy was observed.

The isolated enteric cyst presented here is noteworthy because of its atypical location (anterior mediastinum) and ambiguous CT features. The cyst structure and morphology are similar to those of mature bowel. Hence enteric cyst should be considered in the differential diagnosis of an anterior mediastinal mass, in addition to cystic teratoma and lymphoma. Complete resection is the mainstay of treatment for most cases of mediastinal mass. Even though the cyst reported here was extremely large, the patient underwent resection of the mass with video-assisted thoracoscopic surgery without complication.

References

1. Iglesias Sentis M, Belda Sanchis J, Gimferrer Garolera JM, Catalan Biela M, Rubio Garay M, Ramirez Ruz J. [mediastinal enteric cyst: nusual clinical presentation and histopathology]. Arch Bronconeumol 2004;40:185–7.
2. Sharma S, Nezakatgoo N, Sreenivasan P, Vanatta J, Jabbour N. Foregut cystic developmental malformation: new taxonomy and classification–unifying embryopathological concepts. Indian J Pathol Microbiol 2009;52:461–72.
3. Suzuki K, Koyama S, Yamada S, Kawabata Y. Adenocarcinoma arising in a mediastinal enteric cyst. Intern Med 2007;46:781–4.
4. Ermis B, Yildirim M, Numanoglu V, Er E, Bektas S. Mediastinal enteric cyst: an unusual cause of neonatal respiratory distress. J Pediatr 2010;157:692.
5. Dubrava J, Koren J, Pospisilova V. Mediastinal foregut duplication cyst of enteric type containing a persistent thymus, imitating a pericardial cyst. Bratisl Lek Listy 2013;114:480–3.
6. Carr MJ, Deiraniya AK, Judd PA. Mediastinal cyst containing mural pancreatic tissue. Thorax 1977;32:512–6.