Bronchogenic Cyst in the Intradiaphragmatic Location

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ABSTRACT

Bronchogenic cysts are congenital foregut malformations thought to develop due to abnormal budding of tracheal diverticulum and proximal bronchial structures during embryologic development. The cyst is lined by ciliated pseudostratified columnar epithelium and the wall contains cartilage and layers of smooth muscle. These lesions most commonly are seen in the mediastinum, lung, or pleural spaces. The intradiaphragmatic location of the bronchogenic cyst rarely has been reported in the literature. We report the clinical presentation and computed tomography and magnetic resonance imaging findings in a pediatric patient who presented with left-sided chest pain and was found to have a mass in the region of the diaphragm.

INTRODUCTION

Bronchogenic cysts are developmental foregut malformations and most commonly are found in the mediastinum close to the tracheobronchial tree. Intradiaphragmatic bronchogenic cyst rarely has been reported. We report an unusual case of bronchogenic cyst in a pediatric patient who presented with left-sided chest pain.

CASE PRESENTATION

A 13-year-old boy was referred to our hospital with persistent left lower back pain. His history was significant for having been struck by a motor vehicle approximately 1 year earlier. He sustained bruising on the right chest wall due to the impact, and landed on his left side. Imaging at the time of his initial trauma included radiographs of the chest and cervical, thoracic, and lumbar spine. These were all normal. His pain had been constant and was aggravated by deep inspiration. On examination, he was noted to have mild scoliosis of the dorsolumbar spine with tenderness in the lower thoracic area along the left back. No cardiopulmonary or chest wall abnormality was evident on clinical examination. Due to persistent symptoms for 1 year and no relief with physical therapy and chiropractor treatments, cross-sectional imaging was ordered by his primary care physician. A computed tomography (CT) of the chest abdomen was performed that showed a well-defined ovoid hypoattenuating lesion (30-40 HU) centered in the left crus of the diaphragm adjacent to the descending thoracic aorta and fundus of the stomach (Figure 1). There was no calcification or post-contrast enhancement within this lesion. The differential diagnosis included sequestration, ganglioneuroma, or foregut duplication cyst. The patient underwent magnetic resonance imaging (MRI) of the spine for further characterization and evaluation of intraspinal extension of this lesion. MRI revealed mild scoliosis in the dorsolumbar vertebrae with normal appearance of the spinal cord. In addition, there was a well-defined, homogeneous 4.8 x 3 cm, non-enhancing T1 hypointense lesion (Figure 2a & 2b) and T2 hyperintense lesion (Figure 2c) centered in the left crus of the diaphragm. The lesion appeared cystic with a thin wall and small posterior septation. Based on these findings, a foregut duplication cyst was considered the most likely possibility.

The patient underwent a thorocoscopic excision of the cystic lesion (Figure 3). It was covered by diaphragmatic muscle fibers. The pleural surface of the diaphragm was scored and the muscle was cut to expose the cyst, which was filled with mucus. The diaphragmatic defect was closed with interrupted sutures. Notably, the inferior aspect (abdominal surface) of the diaphragm remained intact. The excised cyst was collapsed, measuring approximately 2.4 x 2.0 cm. Microscopically, the cyst was lined by ciliated pseudostratified columnar epithelium (Figure 4). The cyst wall contained lobules of seromucous secretory units, thin layers of smooth muscle, and islands of cartilage. These findings confirmed the diagnosis of bronchogenic cyst. The post-operative course was uneventful and the patient was discharged on postoperative day 2. He was doing well on his postoperative clinic visit, with resolution of his preoperative back pain.

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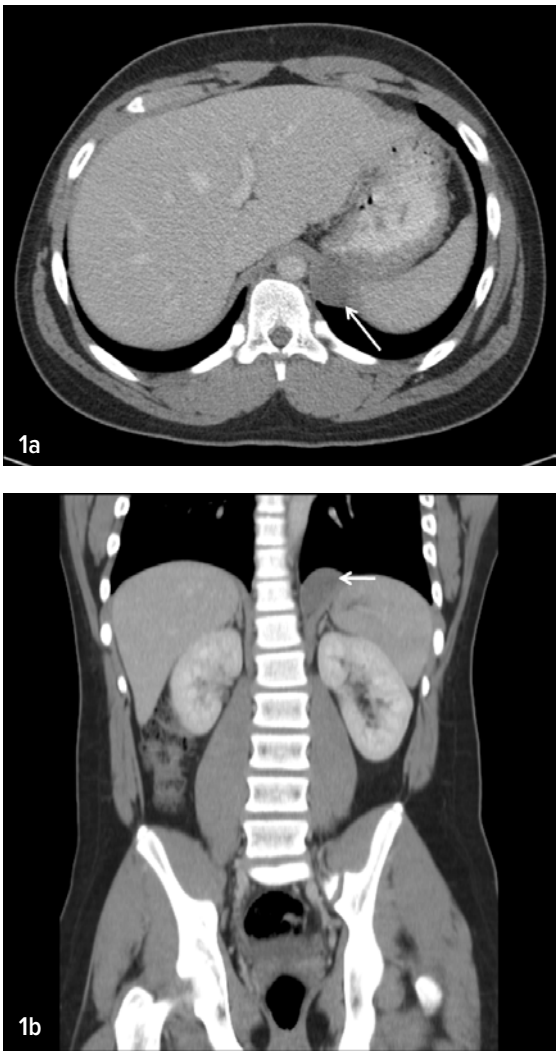
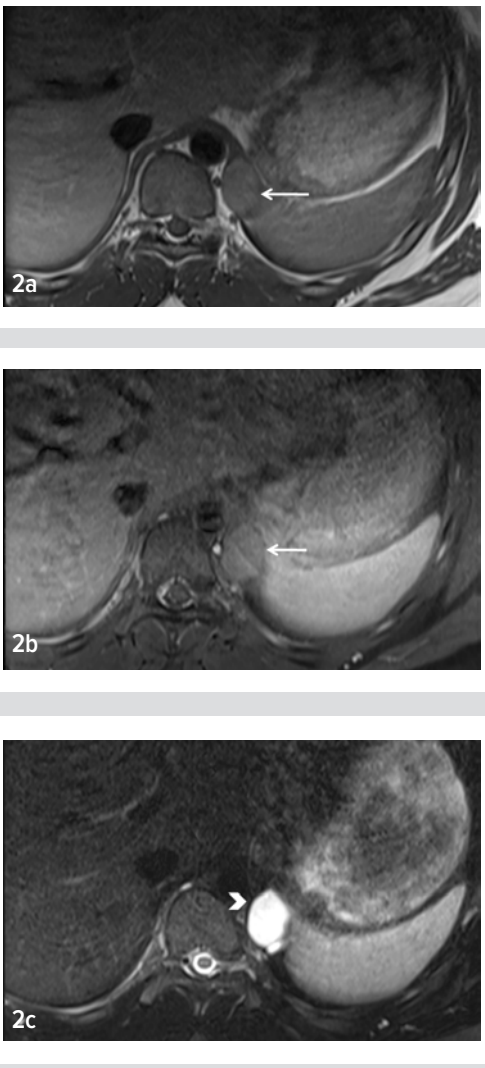
Figure 1. Contrast Enhanced CT of Abdomen



Axial image (1a) and coronal image (1b) show an ovoid hypoattenuating non-enhancing lesion (arrow) splitting the left crus of the diaphragm and abutting the stomach.

Figure 2. Lesions in the Left Crus of the Diaphragm



T1 axial image (2a) and post contrast T1 axial image (2b) show non-enhancing isointense lesion splitting the leaves of left crus diaphragm (arrow). The cyst is hyperintense (arrow head) on the T2 image (2c) and demonstrates a small posterior septation.

DISCUSSION

Brochogenic cysts arise from abnormal budding of the tracheobronchial tree during the 26th to 40th day of gestation.[1] They are lined by respiratory epithelium that enlarges due to accumulation of mucus. These cysts also may contain air if they have communication with the tracheobronchial tree.[1] They usually are found in the mediastinum in 85% patients, and 79% occur in middle mediastinum.[1] Mediastinal bronchogenic cysts in newborns and infants can cause respiratory distress due to compression of the airway, and may require surgical resection.[2] Bronchogenic cysts can be associated with other congenital pulmonary malformations like sequestration or lobar emphysema.[3] They also may be found in the lung, pleura, retroperitoneum, and neck.

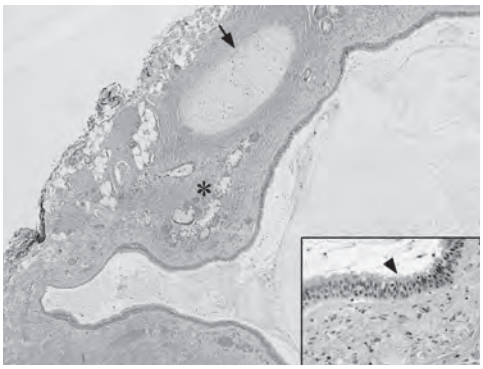
Bronchogenic cysts located within the diaphragm are rare, accounting for less than 30 cases in the English literature. Almost all of these were adults at presentation.[4-6] In a review of 68 patients with bronchogenic cysts, McAdams, et al[1] found only 2 patients with a cyst in the intradiaphragmatic location, both of whom were adults. The only pediatric patient with an intradiaphragmatic bronchogenic cyst was reported by Elemen, et al.[7] They reported a 19-month-old girl who presented with fevers. A chest CT showed a cystic lesion that appeared to be located on segment VIII of the liver. Surgical excision revealed a bronchogenic cyst of the right diaphragm.

Pain was the most common presenting symptom in patients with bronchogenic cyst in most series.[1] However, other presenting symptoms may include fever (due to infection) and symptoms ascribable to pressure on adjacent structures. We believe that the

Figure 3. Thorocoscopic Excision of the Cystic Lesion

Intraoperative image shows the cyst (arrow) within the left diaphragm.

Figure 4. Microphotographs of the Excised Lesion



The cyst is lined by pseudostratified columnar epithelium. The wall shows lobules of seromucous glands (asterisk), thin layers of smooth muscle and islands of cartilage (arrow). Inset: The columnar cells are ciliated (arrowhead). Hematoxylin and eosin stain. Original magnification x50, inset x400.

chronic back pain in our patient was at least in part related to the bronchogenic cyst. It was brought to clinical attention due to trauma. However, trauma did not have any causal relationship to the development of the bronchogenic cyst.

The CT and MRI findings of bronchogenic cyst have been well described in the literature.[1] They are usually sharply marginated with soft tissue or water attenuation, with cystic characteristics. Some bronchogenic cysts may have soft tissue attenuation, and contrast enhanced CT or MRI may help in distinguishing cystic from solid lesion.[1] Ten percent of bronchogenic cysts can have calcification. In our patient, the lesion had attenuation of 30-40 HU, and showed no enhancement or calcification. On MRI, the lesion was hyper-intense on T2W imaging and did not show any restricted diffusion or post contrast enhancement consistent with cystic lesion. Both CT and MRI demonstrated that the lesion was centered in the diaphragm and appeared to split the crus. Apart from bronchogenic cyst, the differential diagnosis for cystic lesions of the diaphragm include gastrointestinal duplication cyst, cystic pulmonary sequestration, cystic teratoma, mesothelium lined cyst, posttraumatic cyst, or hydatid cyst.[5]

The management of diaphragmatic cyst is surgical excision,[3-6] which establishes the diagnosis and relieves any associated symptoms. Malignant transformation of bronchogenic cysts has been reported.[8] Our patient underwent a thoracoscopic excision of the cyst, which considerably shortened his hospital stay and accelerated his return to full function.

CONCLUSION

Intradiaphragmatic bronchogenic cyst presenting in pediatric patients with low back pain is rare. Cross-section imaging (CT/MRI) is required when clinical examination and radiographs are unremarkable and the patient's symptoms persist. It should be considered in the differential diagnosis for any cystic lesion of the diaphragm. In this case, the presence of splitting of the leaves of crura by the lesion helped to localize it to diaphragm. Absence of post-contrast enhancement on CT and MRI and lack of restricted diffusion suggested the cystic nature of the lesion. MRI helps to evaluate for the presence of intra spinal extension. Surgical resection is the treatment of choice.

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