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Case Report

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Atypically located bronchogenic cyst causing invagination

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Abstract

Bronchogenic cysts are formed due to abnormal budding from the bronchial tree and contain cuboidal ciliated epithelium, smooth muscle fibers, submucosal bronchial glands, or cartilage. They are most commonly localized in the lung parenchyma and mediastinum. In the literature, there are reports of very rare atypically located bronchogenic cysts found in the subcutaneous tissue, cervical region, stomach, diaphragm, cardiac septum, retroperitoneum, and abdomen. However, ileal localization of bronchogenic cysts is extremely rare. Although bronchogenic cysts can remain asymptomatic for years, most eventually become symptomatic over time due to compression of surrounding tissues, hemorrhage, or infection. Among the reported symptoms, there has been no documented case of a bronchogenic cyst presenting with intussusception. Herein, we aimed to present a case of atypically located bronchogenic cyst causing ileal intussusception in a 30 month-old male patient. An atypically located bronchoscopic cyst should be considered in cases of intussusception.

Keywords: Bronchogenic cyst, congenital anomalies, ileal intussusception.

Bronchogenic cysts are congenital lesions that develop due to abnormal budding of the embryonic bronchial tree.[1] Their estimated incidence is approximately 1 in 42,000 to 68,000 hospital admissions.[2] Histologically, these cysts may contain cuboidal ciliated epithelium, smooth muscle fibers, submucosal bronchial glands, or cartilage.[1] They are most commonly located in the lung parenchyma.[2,3]

Although bronchogenic cysts most commonly occur in the mediastinum and pulmonary regions, several case reports have documented their occurrence in atypical locations, such as the subcutaneous tissue, cervical region, stomach, diaphragm, cardiac septum, retroperitoneum, and abdominal cavity.[4-10] Nevertheless, ileal localization of bronchogenic cysts remains exceedingly rare.

Although bronchogenic cysts can remain asymptomatic for prolonged periods, they may become symptomatic as a result of compression of adjacent structures, hemorrhage, or infection. To date, there have been no reported cases in the literature of bronchogenic cysts presenting with intussusception. Therefore, we reported this case of intussusception caused by an atypically located bronchogenic cyst.

CASE REPORT

A 2.5-year-old male patient presented to the emergency department with complaints of abdominal pain, nausea, and vomiting that began earlier that day. Abdominal ultrasonography revealed an invaginated bowel loop measuring 35 mm in length and 26 mm in thickness in the right lower quadrant. These findings were interpreted as ileocecal intussusception.

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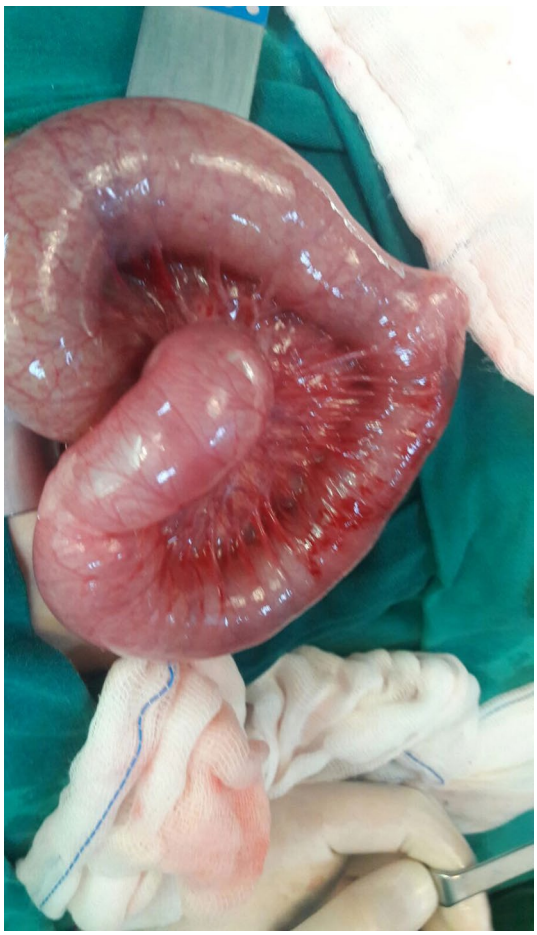

Figure 1. Lead point lesion on the small intestine antimesenteric surface (bronchogenic cysts).

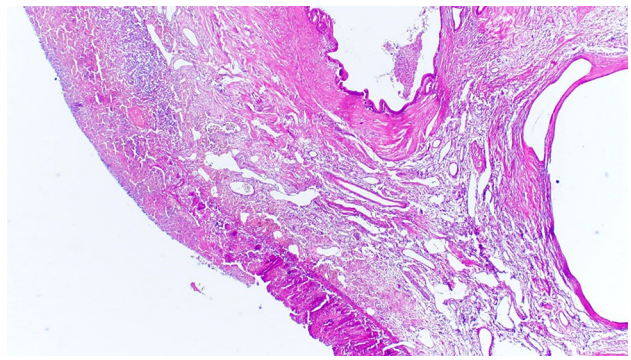

Figure 2. Ulcerated ileum with cystic lesion under the epithelium (H&E, ×100).

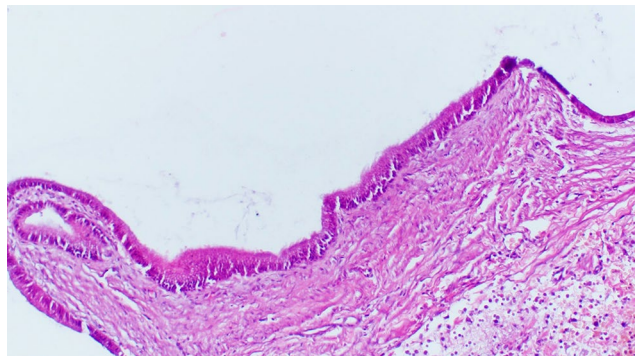

Figure 3. Cystic lesion lined by ciliary epithelium (H&E, ×100).

A saline enema was performed under ultrasonographic guidance to reduce the intussusception, but the attempt was unsuccessful. Consequently, exploratory laparotomy was performed. During surgery, a 15-cm segment of ileoileal intussusception was identified and manually reduced. A lesion acting as the lead point was found on the antimesenteric surface of the bowel, located 15 cm proximal to the cecum (Figure 1). The affected segment, along with adjacent proximal and distal bowel loops, was resected, and primary end-to-end anastomosis was performed.

The postoperative course was uneventful, and the patient was discharged on the seventh day. Histopathological examination of the resected specimen revealed multiple cystic structures beneath the ulcerated ileal epithelium, with no connection to the surrounding mucosa (Figures 2 and 3). These cysts were lined by ciliated epithelium resembling bronchial tissue and were diagnosed as bronchogenic cysts. A written informed consent was obtained from the parent of the patient.

DISCUSSION

The diagnosis of intussusception is typically based on clinical findings and confirmed via abdominal ultrasonography or barium enema. Ultrasonography has been reported to have a diagnostic sensitivity of 98 to 100% in detecting intussusception.[11,12]

Nonsurgical enema-based reduction techniques generally have a high success rate (over 90%). In our case, although a saline enema was attempted under ultrasonographic guidance, it failed to achieve reduction.

Surgical intervention is crucial not only for achieving reduction but also for detecting

possible pathological lead points and preventing complications. In approximately half of pediatric intussusception cases requiring surgery, a pathological lead point is identified.[13,14] In our case, the lead point was discovered during surgery following failed enema reduction.

Bronchogenic cysts are generally benign congenital anomalies most commonly located in the lung parenchyma and posterior mediastinum.[15,16] Although the pathophysiology is not fully understood, it is believed that bronchogenic cysts result from malformations occurring between the third and seventh weeks of embryonic development, involving abnormal communication between the primitive foregut and the developing lung bud.[17] This malformation is thought to arise from the failed separation of the lung bud from the esophagus and trachea, followed by migration into the thoracic or abdominal cavity.[17]

Although they are most frequently found in the posterior mediastinum, bronchogenic cysts have also been reported in the stomach, retroperitoneum, pancreatic tail, spleen, and left adrenal gland.[18] They are most commonly found in the lung parenchyma, with a reported incidence of 20%.[19,20]

Histopathologically, the distinction between bronchogenic cysts and other foregut-derived congenital malformations, such as duplication cysts, is made based on the epithelial lining and the composition of the cyst wall, including the presence of smooth muscle, cartilage, and bronchial glands.[21]

Extrathoracic bronchogenic cysts were reviewed by Casagrande and Pederiva[18] in 20 different studies, with most reported cases located in the retroperitoneal area or near the upper pole of the left kidney.[22] Some reports have also suggested that bronchogenic cysts can rarely be found in the ileum.[23,24]

In our case, the cyst was located adjacent to the ileum, a localization not previously reported in this age group. Furthermore, to our knowledge, no report of a bronchogenic cyst causing intussusception has been reported. Due to this unusual location, the definitive diagnosis was established only after histopathological examination.

One hypothesis in the literature for such ectopic locations suggests abnormal migration of the tracheobronchial bud into the abdominal cavity during early embryonic development, before the diaphragm has fully formed. During this period, the thoracic and abdominal cavities remain connected, allowing for the potential displacement of bronchial tissue into the abdominal region.[9,25] The intra-abdominal bronchogenic cyst identified in our case supports this embryologic theory.

Most bronchogenic cysts are asymptomatic during childhood, particularly those that are small in size. Symptomatic cases in the literature are most commonly associated with infection, perforation, or malignant transformation.[18,26] Therefore, complete surgical excision is essential in both pediatric and adult patients to prevent progression or malignancy. In our case, no evidence of malignancy or lymphadenopathy was observed in the excised tissue. However, malignant transformation in later decades has been reported in the literature.[26]

In conclusion, in pediatric cases of intussusception, the possibility of a pathological lead point should always be considered. Although extremely rare, bronchogenic cysts should be included in the differential diagnosis of such lead points. This case is noteworthy as one of the first reported instances in the literature of an ileal bronchogenic cyst causing intussusception in a young child. We believe that in addition to more common lead points, such as Meckel's diverticulum, duplication cysts, and malignancies, bronchogenic cysts should also be kept in mind as a potential cause.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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