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WORD COUNT 2633
TIME SUBMITTED 28-SEP-2022 09:54AM
PAPER ID 91014785

Name of Journal: *World Journal of Clinical Cases*

Manuscript NO: 78737

Manuscript Type: CASE REPORT

Abdominal bronchogenic cyst: A rare case report

Li C *et al*. Abdominal bronchogenic cyst

Abstract

BACKGROUND

Bronchogenic cysts are cystic masses caused by the congenital abnormal development of the respiratory system, which usually occurs in the pulmonary parenchyma or mediastinum.

CASE SUMMARY

A rare case of a bronchogenic cyst discovered in the abdominal cavity of a 35-year-old male was reported. Physical examination found a space-occupying lesion in the patient's abdomen for four days. Laparoscopic exploration found the cyst tightly adhered to the stomach and its peripheral blood vessels; therefore, intraoperative laparotomy was performed. The cystic mass was resected *en bloc* with an endo-GIA. The final postoperative pathological diagnosis confirmed an abdominal bronchogenic cyst.

CONCLUSION

This is a rare case of a bronchogenic cyst that was discovered within the abdominal cavity of a male patient. The cyst is easily confused with or misdiagnosed as other lesions. Therefore, it is necessary to distinguish abdominal bronchogenic cyst from gastrointestinal stromal tumor, Meckel's diverticulum, enteric duplication cyst, or lymphangioma. Though computer tomography scan and a magnetic resonance imaging examinations were the primary diagnostic approaches, endoscopic ultrasound-guided

fine-needle aspiration could assist the clarification of the cytologic diagnosis or histopathologic diagnosis before the surgery.

Key Words: Bronchogenic cyst; Abdominal cavity; Endoscopic ultrasound-guided fine-needle aspiration; Case report

INTRODUCTION

Bronchogenic cysts are cystic masses caused by the congenital abnormal development of the respiratory system, which usually occurs in the pulmonary parenchyma or mediastinum. Abdominal bronchogenic cysts are rarely documented. We report a rare case of an ectopic bronchogenic cyst within the abdominal cavity of a 35-year-old male patient.

CASE PRESENTATION

Chief complaints

Physical examination revealed an abdominal space-occupying lesion in a 35-year-old male patient for four days.

History of present illness

Four days ago, the patient presented to our hospital for physical examination. The physical examination discovered a space-occupying lesion in his abdomen.

History of past illness

A space-occupying lesion was found in the patient's abdomen after the physical examination. The patient was admitted to our department for further surgical treatment. Abdominal computed tomography (CT) and enhanced CT revealed a hepatogastric space-occupying lesion. Abdomystic lymphangioma was initially

suspected. There were no any complaints of abdominal pain, fever, nausea, or vomiting after admission.

Personal and family history

The patient once underwent laparoscopic argon knife surgery for gastric, cardia, and colon polyps. Past medical history showed that the patient was a hepatitis B surface antigen (HBsAg) carrier. Family history was denied. Both Infectious and genetic diseases were also denied.

Physical examination

The following are the patient's vital signs: Body temperature, 36.5℃; heart rate, 69 beats per min; respiratory rate, 16 breaths per min; blood pressure, 110/70 mmHg. Cardiopulmonary examination was normal. The whole abdomen touched soft, without tenderness and rebound tenderness. Examination of liver, gallbladder, spleen and both kidneys revealed no abnormalities. Bowel sounds 4 times per min.

Laboratory examinations

No abnormality was found in routine blood, urine and excrement analyses, hepatonephric function, and blood coagulation tests. Blood transfusion-related tests: HBsAg was positive (+); hepatitis B core antibody determination was positive (+); hepatitis B e antibody determination was positive (+); hepatitis C, treponema pallidum and human immunodeficiency virus antibodies were all negative (-).

Imaging examinations

Chest CT scan and B-ultrasonic examination of liver, gallbladder, pancreas, spleen and double kidneys revealed no abnormality. Abdominal CT and an enhanced CT scan revealed a hepatogastric space-occupying lesion, which was considered an abdomystic lymphangioma (Figure 1).

FINAL DIAGNOSIS

The final diagnosis was ectopic bronchogenic cyst within the abdominal cavity.

TREATMENT

The patient underwent laparoscopic exploration under general anesthesia with endotracheal intubation. Abdominoscope discovered the cyst was located between the lower left diaphragm, aorta abdominalis, left gastric vessel, splenic artery, and pancreas. The laparoscopic exploration found the cyst tightly adhered to the stomach and its peripheral blood vessels, so the surgery was conversed to laparotomy, and the cystic mass was resected *en bloc* by Endo-GIA stapler (Figure 2). The surgery went smoothly. The final postoperative pathological diagnosis confirmed the abdominal bronchogenic cyst (Figure 3).

OUTCOME AND FOLLOW-UP

The patient recovered very well upon follow-up three months after the surgery.

DISCUSSION

CT and MRI are generally considered the major diagnostic approaches of abdominal masses. Most of the bronchogenic lesions are reported to be cystic ones, and only a few are solid. Therefore, it is valuable to be distinguished from gastrointestinal stromal tumors, Meckel's diverticulum, cystic intestinal duplication and lymphangioma. CT and MRI could help to identify the imaging features of the lesions, and endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) could assist the surgeons to make clear the tissue source of the lesions and to confirm the cyst benign or malignant.

The exact pathogenesis of bronchogenic cysts remains unknown. It is possibly related to embryonic development. During the embryonic development, the epithelium of the respiratory tract is separated from the tracheobronchial tree, migrates from the previously developed site to distal sites, and gradually expands. Its increased internal mucus secretion which cannot be discharged results in a mucus-containing cyst

consisting of bronchial tissue comprising its walls. The disease is categorized into three types according to the pathogenic location: mediastinal, intrapulmonary, and ectopic. The mediastinal type is more common, the intrapulmonary type is less common, while the ectopic type is extremely rare, occurring in intracranial cavities, sublingual cavities, diaphragm, and the abdominal cavity[1]. Abdominal bronchogenic cysts, including abdominal and retroperitoneal cysts, are extremely rare. There are few documented cases of bronchial cysts within the abdominal cavity, most of which describe lesions identified on the side of the retroperitoneal region, while cases of lesions attached to the stomach were extremely unusual[2]. No specific clinical symptoms are presented when the cyst is small, so abdominal bronchogenic cysts are difficult to diagnose preoperatively and are usually discovered through physical examinations, so they are more easily misdiagnosed as an abdominal space-occupying lesion. Imaging of abdominal bronchogenic cysts reveals circular or elliptical cystic spaces occupying primarily single cavities with well-defined margins. The long axis of the cyst in the abdominal tissue space is consistent with the long axis of the tissues, which may compress the adjacent tissues. No enhancement is found on the contrast-enhanced CT, except the thickened cystic wall showing annular enhancement. CT and MRI can provide more valuable imaging characteristics of the lesion, while the EUS-FNA is recognized as an effective technique for obtaining tissue samples[3-6], which could cause minimal injury for the patients. This technique can assist the surgeons to clarify the tissue source of the tumor and determine the benign or malignant nature of the tumor. Therefore, it is quite valuable for the surgeons. This technique is adaptable to some abdominal, retroperitoneal, and mediastinal lesions, which helps to make a definite cytodiagnosis or histopathological diagnosis possible before surgery[7]. However, if tissue necrosis or hemorrhage occurs, the pathological results might be affected. Different diagnosis of the abdominal bronchogenic cysts includes gastrointestinal stromal tumor, Meckel's diverticulum, intestinal duplication cyst, and lymphangioma, *etc*[8]. We consider the preoperative imaging examination an important approach, which will differentiate the abdominal bronchogenic cysts from gastrointestinal stromal

tumor, Meckel's diverticulum, intestinal duplication cyst and lymphangioma, *etc.* The image of the low-grade malignant gastrointestinal stromal tumors is generally less than 5 cm in diameter, with a regular well-circumscribed shape, minimally compressing the adjacent tissues. Malignant tumors are generally larger than 5 cm, with undefined margins, uneven density, and are normally accompanied by necrosis, bleeding, calcification, cystic change, and invasion into the adjacent tissues. Severe necrosis can communicate with the intestinal cavity to form a gas-liquid surface, indicating symptoms of a pseudo-intestinal cavity. However, having few signs of intestinal obstruction[9]. The CT value of malignant gastrointestinal stromal tumors increases significantly, surrounded by garland-shaped or irregular patchy enhancements. The blood supply in most lesions of gastrointestinal stromal tumors is significantly enhanced, and CT values of the venous phase are higher than that of the arterial phase. A higher expression of CD117 and DOG-1 can be sensitive markers for small intestine stroma tumor (SIST) or gastrointestinal stromal tumor. Tumor diameter 5.3 cm or higher and nuclear division number > 5/50 can be independent risk factors to predict postoperative adverse outcome for SIST patients[10].

Meckel's diverticulum is a distal ileum diverticulum formed by an incomplete vitelline canal, which communicates with the ileum during embryonic development. The most usual complications of Meckel's diverticulum include bleeding and obstruction. Bleeding is a frequent childhood life-threatening complication, while intestinal obstruction is a common complication in adulthood[11]. Routine barium enema retrograde ileography is performed, indicating that a pleated sac filled with contrast medium has adhered to the mesenteric margin of the small intestine. New diagnostic methods such as capsule endoscopy, double-balloon enteroscopy and MR enterography have emerged in recent years[12]. Intestinal duplication cysts (enteric duplication cysts) are rare congenital gastrointestinal malformations. According to medical reports, the incidence of enteric duplication cysts is 1:4500 births[13], and their aetiology remains unclear. Due to the different sizes, locations, types, and mucosal patterns of the cysts, their clinical presentations are also varied[13]. Intestinal duplication cyst occurs more

commonly in infants and children under two years of age and often results in abdominal pain, intestinal obstruction, hematochezia, and peritonitis. Intestinal duplication cysts in adulthood are the potential risks for malignant transformation[13]. Abdominal ultrasonography is the most commonly used diagnostic approach due to its more frequent occurrence in young children. The ultrasound imaging show that the intestinal duplication cyst consists of five layers: mucosal, submucosa, mucosal muscularis, muscularis propria and serosal layer[14-17]. For imaging diagnosis, enteric duplication cysts could be confirmed *via* mucosal patterns and smooth muscle layers, which must adhere to the intestinal fistula[18,19]. Other cystic lesions in the abdomen like the omental cyst or mesenteric cyst, do not demonstrate this characteristic multi-layered wall[17]. Abdominal cystic lymphangioma, usually congenital, is more common in children and rare in adults. It often develops along the abdominal space. When the lesion is small, there is no apparent compression on the adjacent tissues. As the lesion increases, it is often distributed along the omentum, retroperitoneal cavity, and mesentery, demonstrating the characteristics of crawling growth in the crevices. CT scans show a well-defined thin-walled cystic lesion with multilocular capsules and visible partitions. The partitions and wall can be strengthened without wall nodules. Laparoscopic surgery is the most used treatment for abdominal bronchogenic cysts. If surgical operation is not performed, infection, bleeding, and malignant transformation may occur[19].

CONCLUSION

As previously reported, it is necessary to thoroughly inquire the patient of the onset age, the common symptoms, and clinical presentations of the disease. As for the abdominal space-occupying lesions that have not been clearly diagnosed, apart from the preoperative abdominal CT and MRI examinations, EUS-FNA is proposed to be performed as a viable diagnostic procedure. However, EUS-FNA has not been widely applied as a useful diagnostic technique in most hospitals. CT and MRI modalities are helpful to better perceive the imaging characteristics of the mass, while EUS-FNA may

help the surgeons to confirm both the source of the tissues and to define the nature of the cyst. Intraoperatively, if the cyst is found closely adhered to the adjacent organs, tissues, and blood vessels, damage to the capsule wall should be avoided to ensure a complete surgical resection to reduce the recurrence rate. Postoperative follow-up is recommended. Routine postoperative histopathological examination is generally required to clarify the tissue source and to determine whether the cyst is benign or malignant. Resections should be performed if the cyst is confirmed malignant.

ACKNOWLEDGEMENTS

The authors would like to express special gratitude to Professor Liang Zhou, surgeons Wei-Hua Huang and Wei-Wei Fan of Department of General Surgery, Xi'an Gaoxin Hospital for their support to publish this article.

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Figures 1 Arterial, venous, and delayed phases, respectively. A: Computed tomography (CT) value was 44HU; B: CT value was 39HU; C: CT value was 35HU. The lesion had a well-defined margin and was adhered to the posterior wall of the gastric cardia and the inner wall of the gastric fundus, adjacent to the gastric mucosal lining with visible surrounding fat space.

Figure 2 Resected section of the cystic mass postoperatively.

Figure 3 The cystic wall lined with columnar epithelium with cartilage and mucous glands. Hematoxylin and eosin stain, 10 × 4.

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