



Surgery for Intradiaphragmatic Bronchogenic Cysts: Report of 4 Cases and Literature Review

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Abstract

Bronchogenic Cysts (BC) are congenital lesions found mostly around the tracheobronchial tree and in the mediastinum, but rarely in the diaphragm. In here, we report 4 cases of patients with intradiaphragmatic Bronchogenic Cysts (BCs) at our hospital and review the related literature published in English. We characterized the clinical course, preoperative diagnoses, Computed Tomography (CT) imaging, surgical approaches to resection, extent of resection, and pathological characteristics of each case. We focused mainly on the surgical procedure to treat the intradiaphragmatic BCs.

Keywords: Bronchogenic cysts; Intradiaphragmatic; Diaphragm; Surgery; Surgical approaches

Introduction

Bronchogenic Cysts (BC) are cystic masses caused by congenital abnormal development. They can be divided into pulmonary, mediastinal, and ectopic types according to their location. Intradiaphragmatic BCs are rare. We present 4 cases of patients with intradiaphragmatic BCs who attended our hospital, we collected data on their clinical course, preoperative diagnosis, imaging features, surgical approaches to resection, extent of resection, and pathological characteristics; in addition, we reviewed the literature to compare these cases with others.

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Received Date: 23 Nov 2020

Accepted Date: 06 Jan 2021

Published Date: 12 Jan 2021

Citation:

Xiao X-X, Chang R-M, Gao Y, Chen Y-D, Xia X-F, Zhang W-X. Surgery for Intradiaphragmatic Bronchogenic Cysts: Report of 4 Cases and Literature Review. *Clin Surg.* 2021; 6: 3023.

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Case Presentation

We encountered 4 Asian patients (3 men and 1 woman) with intradiaphragmatic BCs in our hospital. Their average age was 50 years, and they all lacked trauma histories. The men had all smoked. Only one of the men was asymptomatic. The preoperative diagnoses were mainly based on Computed Tomography (CT) imaging results. Figure 1 compares thoracic CT findings for each patient and Figure 2 displayed pathological characteristics. Table 1 shows the baseline characteristics of the patients. Table 2 lists surgical approaches, intra-operative details, and post-operative recovery days. One patient had to undergo a second operation because of coronary artery bleeding on the

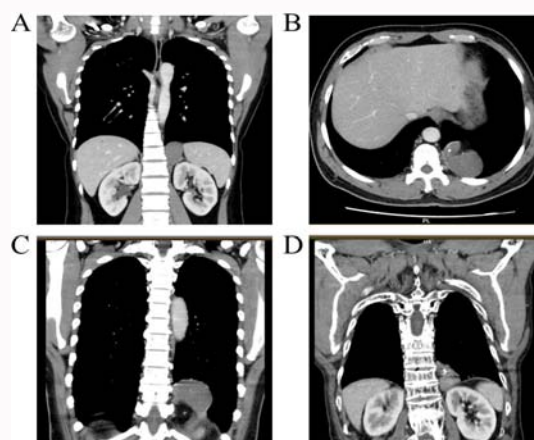


Figure 1: Thoracic CTs of patients. A) case 1, 51 years; B) case 2, 38years; C) case 3, 55 years; D) case 4, 56 years. CT showing lesions as soft tissue hypo-enhancing masses adhering to the left diaphragm, calcifications were present in four patients (combined linear and nodular).

Table 1: Baseline patients' characteristics.

Case number	Age (year)	Gender	Past history	Personally history	Prevalent symptoms	Preoperative diagnosis
1	51	Male	None	Smoking	None	Mediastinal neurogenic tumors
2	38	Female	None	None	Chest pain	Diaphragmatic occupying
3	55	Male	None	Smoking	Chest tightness and cough	Left lung occupying
4	56	Male	Hypertension, gout, hepatitis B	Smoking	Chest tightness and cough	Diaphragmatic occupying

Table 2: Surgical operation-related details.

Case number	Surgical approach	Operation time (min)	Diaphragmatic management	Intraoperative bleeding (ml)	Complications	Hospital stay after operation (days)
1	Thoracoscopy	40	Primary closure	30	No	4
2	Thoracoscopy	38	Primary closure	20	No	4
3	Thoracoscopy	40	Primary closure	30	Yes	11
4	Thoracotomy	70	Gore-Tex patch reconstruction	100	No	3

second postoperative day and he was discharged after 11 days. The pathology reports showed benign ciliated respiratory epithelium, cartilage, and sero-mucinous glands.

Methods

We performed a combined MEDLINE, PubMed, ScienceDirect, and SCOPUS Database search for “bronchogenic cyst” AND “diaphragm” without date limitations. We selected all the case reports in English we found for the analysis and excluded articles in other languages. The cases selected included CT findings, pathology descriptions, surgical approaches, and diaphragmatic management. We collected patient’s data including age, gender, symptoms, past history, CT findings, surgical approaches, diaphragmatic management, length of hospital stay after the operations, and post-operative complications. We excluded case reports lacking pertinent data from each particular analysis (such as the analysis of the operative approach or diaphragmatic management). The ethics committee of Xiangya Hospital (Central South University) approved this study. All procedures involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Results

We found 14cases of intradiaphragmatic bronchogenic cysts published in English from 1957 to 2019. The age at presentation ranged from 19 months to 74 years. Including the 4 cases in our hospital, we analyzed 18 cases. The mean age of all the patients was 37.3 years. Most of them were men (61.1%). Almost one quarter of them (22.2% or 4/18) had no symptoms. Table 3 details the symptoms of the rest

Table 3: Clinical course.

Presenting signs and symptoms	Percentage (%)
Pain (chest, abdominal, back)	38.9 (7)
Previous trauma	11.1 (2)
Cough	5.6 (1)
Urination abnormalities	5.6 (1)
Uncomplained fatigue	5.6 (1)
Hemoptysis with unknown reason	5.6 (1)
Fever of unknown reason	5.6 (1)
No symptoms	22.2 (4)

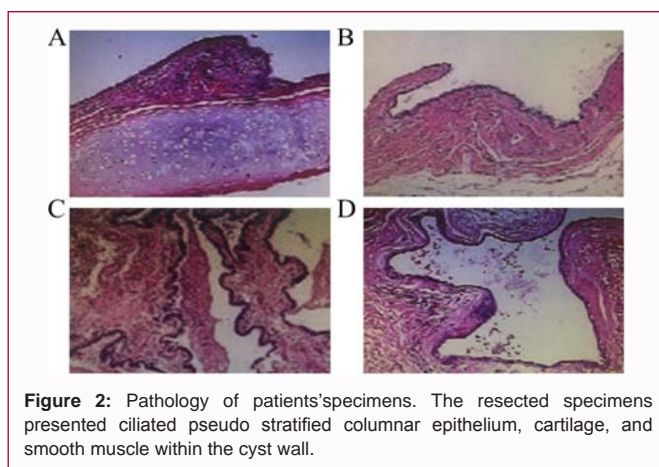


Figure 2: Pathology of patients' specimens. The resected specimens presented ciliated pseudo stratified columnar epithelium, cartilage, and smooth muscle within the cyst wall.

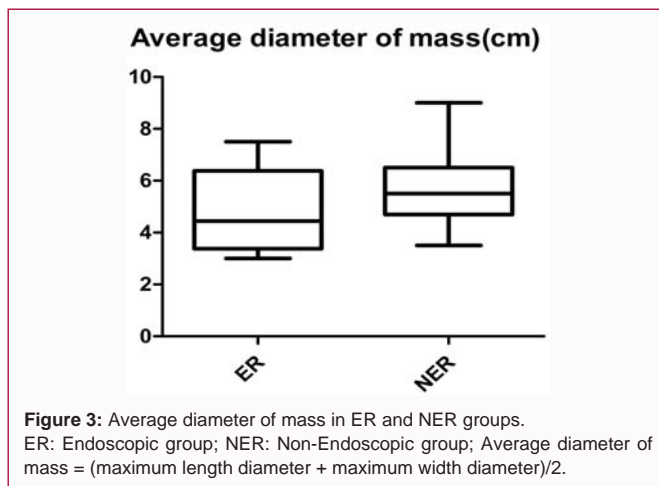


Figure 3: Average diameter of mass in ER and NER groups. ER: Endoscopic group; NER: Non-Endoscopic group; Average diameter of mass = (maximum length diameter + maximum width diameter)/2.

of the patients. The locations of the cysts included 9 (50.0%) in the diaphragm, while others were classified as hepatic, mediastinal, adrenal or lung occupying lesions and one was diagnosed as a diaphragmatic hernia. Only 3 patients (16.7%) were diagnosed as having cysts before the operation, and 2 of them had been detected in MRI findings. All the case descriptions in our analysis included surgical approaches and diaphragmatic management details (Table 4). The procedures included thoracotomies in 6 patients (34.2%), laparotomies in 3(16.7%), thorascopies in 5(27.8%), thoracoabdominal incisions in 2 (11.4%), laparoscopy in 1 (5.6%), and initial thoracoscopy with

Table 4: Review of the cases of intradiaphragmatic bronchogenic cyst in the English literature.

English literature									
First author	Year	Age	Sex	Admission diagnosis	Tumor size	Surgical approach	Diaphragmatic management	MRI	POD
Francois Dagenais [7]	1995	51	Female	Hepatic mass (neurofibroma)	4 × 3 cm	Right posterolateral (10 ribs)	Primary closure	No	N/A
Subramaniam [8]	1996	50	Male	Adrenal gland malignancy tumor	4 × 9 cm	Left thoracoabdominal (11 ribs)	Primary closure (2,0 vicryl)	No	8
Chang-Hsien Liou[9]	2001	34	Male	Posteriormediastinal tumor	5 × 7 cm	Left posterolateral thoracotomy	Primary closure	No	
Fernando Luiz Westphal [10]	2003	32	Male	Diaphragmatic hernia	5 cm defect	Left postero-lateral thoracotomy	Non-absorbable suture stitches anchored in prolene mesh	no	4
Zhong-Cheng Li [11]	2019	16	Female	Diaphragmatic tumor	6.5 × 5.5 cm	Right thoroscopic	Interrupted sutures	No	N/A
LeventElemen [12]	2008	19 months	Girl	Hydatid cyst	4 × 3 cm	Explorative laparotomy(left)	No injury to diaphragmatic	No	2
AS Valero Liñán [13]	2009	26	Male	Left diaphragmatic mass	8 × 4	Midline laparotomy(left)	Part of the diaphragm was excised and directly sutured	No(have ct biopsy)	7
JaeBum Kim [14]	2011	56	Female	Right diaphragm mass	8.5 × 9.5	Right lateral side	7x8 cm defect in the diaphragm, Gore-Tex 2 mm patch after removal of the tumor	No	7
Subramanian Subramanian [15]	2013	13	Male	Foregut duplication cyst	4.8 × 3 cm	Left thoroscopic excision	Interrupted sutures, the inferior aspect (abdominal surface) of the diaphragm remained intact.	Yes	2
Chao Jiang [16]	2013	38	Female	Left crus of the diaphragm mass	5 × 5	Open left thoracotomy	Directly closed by figure-of-eight sutures	No	N/A
Nico P Zu"ge [17]	2008	43	Female	Symptomatic cystic liver tumors	8 × 5 cm	Laparoscopic Resection	Cystic tumor was completely resected including a 1-cm muscle margin from the diaphragm	No	4
Ronnie Mubang [18]	2015	41	Male	Bronchogenic or proteinaceous cyst	4.5 × 5 cm	Left posterolateral serratus	Diaphragm was reconstructed with a synthetic patch	Yes	5
XinGao [19]	2017	22	Male	Left Dumbbell-shaped mass	12.7 × 13.1 cm	exploratory laparotomy	Repaired with interrupted 0 silk suture	No	N/A
Buddington WT [4]	1957	62	Male	Left adrenal gland tumor	6.5 cm	Thoracoabdominal	Repaired with mattress sutures of heavy silk	No	15
Our cases									
Present case		38	Female	Diaphragmatic occupying	4 × 3 cm	Thoracoscopy	No injury to diaphragmatic muscle	No	4
		55	Male	Left lung occupying	3 × 3 cm	Thoracoscopy	No injury to diaphragmatic muscle	No	4
		56	Male	Diaphragmatic occupying	5.4 × 3.9 cm	Thoracotomy	Gore-tex patch reconstruction	No	3
Yuanda CHENG [1]	2018	51	Male	Mediastinal neurofibroma	4.3 × 5.8 cm	Left VATS	No injury	No	11

subsequent thoracotomy in 1 (5.6%). We divided the patients into an endoscopic group and a non-endoscopic group. Table 4 shows the maximum lesion diameters, the average diameters of endoscopic and non-endoscopic groups were 5.4 cm and 6.7 cm, respectively (Figure 3 shows a boxplot with detailed data). We also analyzed differences in terms of hospital length of stay after the surgical procedures between these two groups (Table 4).

Discussion

Intradiaphragmatic bronchogenic cysts are exceedingly rare,

but some practitioners think that bronchogenic cysts may undergo malignant degeneration [1,2]. Surgery is a good choice to treat the lesion. Symptoms include pain (35.7% of patients), cough (10.7%), and dyspnea (7.1%) due to compression or irritation of adjacent structures; while other patients (39.3%) are asymptomatic. The detection of these lesions is mostly incidental during routine physical examinations or studies for other conditions. In our review, we found one patient reported to have developed a diaphragmatic hernia after trauma. The differential diagnosis of intradiaphragmatic bronchogenic cysts depends on the tumor pathology and its

properties. According to their composition, intradiaphragmatic BCs can be misdiagnosed as lung, mediastinal or abdominal occupying masses (especially as adrenal gland tumors). According to their properties, BCs can be misdiagnosed as diaphragmatic tumors, diaphragmatic hernias, hydatid cysts, esophageal diverticula, or neurofibromas. The primary admission diagnoses depend mostly on imaging examinations including chest X-rays, ultrasonography, CT, and Magnetic Resonance (MR) images. In our review, only one patient did not undergo CT scanning [3] and 2 patients had MRI examinations. The accuracy of the preoperative diagnosis was better in patients who underwent MRI (50%, 2/4) than in those who lacked MRIs (0/16). On CT scans, BCs appear as benign-appearing cystic masses, contrast may show a thin peripheral enhancement, and sometimes calcifications [4]. On MRIs, BCs can have low signal intensity on T1-weighted and hyperintensity on T2-weighted images. With the combination of CT and MRI, the diagnosis can be easier and more accurate. Complete surgical excision is the best choice for curing intradiaphragmatic BCs. The surgical approaches include thoracotomy, laparotomy, thoracoabdominal incision, and minimally invasive surgery (thoracoscopic and laparoscopic) according to our literature review. We were able to directly look at the lesions in our patients using endoscopy. For small-diameter cysts, the complete cyst resection can be accomplished through endoscopy and the diaphragm can be simultaneously repaired. According to our review, the average length of hospital stay after the operation for patients who underwent endoscopic procedures was shorter than that of patients who underwent non-endoscopic procedures, which may be explained by the low frequency of postoperative pain and the early respiratory function recovery after the endoscopic procedures [5]. In cases with uncertain diagnosis, the endoscope can be of value before planning the surgical removal of the lesion by an alternative method after its visualization. The pathology of bronchogenic cysts consists of ciliated pseudo stratified columnar epithelial cells, cartilage, and smooth muscle within the cyst wall.

Conclusion

The incidence of intradiaphragmatic bronchogenic cysts is extremely low and difficult to diagnose. A combination of thoracoabdominal CT and MRI can help physicians make a diagnosis. Surgery is the main treatment, our review of the literature suggests that an initial endoscopic exploration can be useful when the diagnosis is unclear, and that the decision to switch to an open surgery should be taken based on the intraoperative conditions.

Funding Sources

This work was supported by the National Natural Science Foundation of China (grant numbers 81401901, 81372515, and 81602027), and by the Key Research and Development Program of Hunan Province (Grant Number: 2016JC2039).

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