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CASE REPORT

Congenital bronchobiliary fistula: A case report and review of the literature

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Abstract

BACKGROUND

Congenital bronchobiliary fistula is a rare developmental abnormality with an abnormal fistula between the respiratory system and biliary tract. The aim of this report is to analyze and summarize the clinical features and experience of diagnosing and treating congenital bronchobiliary fistula (CBBF) occurring in the neonatal period.

CASE SUMMARY

The onset of symptoms was 3 d after birth in our patient with progressive cyanosis and respiratory distress, and a large amount of green fluid was noticed in her respiratory secretion. We performed computed tomography (CT), fiberoptic bronchoscopy, and cholangiography to make a diagnosis, as well as fistulography with a bronchoscope for the first time. These examinations provided us with valuable images to make a correct diagnosis. The fistula was dissected and removed with excellent results. Surgical removal of the fistula was successful, and the baby recovered well and was discharged. She has been followed for 4 mo without any signs of discomfort.

CONCLUSION

The main symptom of CBBF is bile-like sputum. CT, bronchoscopy, fistulography, and intraoperative cholangiography can provide important evidence for diagnosis. Surgical resection of the fistula is the first choice of treatment.

Key words: Congenital bronchobiliary fistula; Neonate; Computed tomography; Bronchoscopy; Fistulography; Case report

Core tip: Congenital bronchobiliary fistula is a rare developmental abnormality with an

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abnormal fistula between the respiratory system and biliary tract. We summarize the clinical data, and review related papers on it. The aim of this report is to analyze and summarize the clinical features and experience of diagnosing and treating congenital bronchobiliary fistula occurring in the neonatal period.

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INTRODUCTION

Congenital bronchobiliary fistula (CBBF) is a rare developmental abnormality with an abnormal fistula between the respiratory system (trachea or bronchus) and biliary tract. Patients with this anomaly may develop symptoms at any time from neonate age to adulthood, but most of them present with signs of apnea, bilious saliva, and choking, within just a few days after birth. Generally, the earlier the symptoms appear, the more serious the condition is. CBBF is often accompanied by biliary tract developmental anomalies, such as biliary atresia. Surgical resection of the fistula is the ultimate choice of treatment. Presently, this anomaly is believed to be caused by abnormal development of the foregut[1-3]. In this paper, we describe a patient with CBBF who showed the symptom of apnea at 3 d after birth, summarize the clinical data, and review the related papers on CBBF.

CASE PRESENTATION

Chief complaints

Intermittent cyanosis for 3 d.

History of present illness

A full-term female baby of 5 d was referred to our hospital because of intermittent cyanosis. She secreted a large amount of yellowish and green saliva and started choking since 3 d of age. Respiratory distress became increasingly severe after admission, so tracheal intubation and a ventilator were applied thereafter. After intubation, yellowish green fluid kept flowing out of the tracheal tube. Gastroesophageal reflux complicated by a tracheoesophageal fistula was considered first, but nasogastric depression produced colorless fluid, which was not in accordance with the aforementioned findings.

Physical examination upon admission

Jaundice.

Laboratory examinations

Total bilirubin 172.4 μmol/L.

Imaging examinations

We performed 3-dimensional (3D) computed tomography (CT) of the baby's chest. Her trachea and esophagus were reconstructed, and an abnormal fistula originated from the right bronchus; the fistula formed down along the esophagus and passed through the diaphragm into the intrahepatic biliary tract at the site of the esophageal hiatus. Gas was noticed in the intrahepatic biliary tract and common hepatic duct. Furthermore, pneumonia was evident in both lungs, especially in the lower lobes (Figure 1A-C). CBBF was diagnosed accordingly.

During the ultrasonographic examination, we discovered an abnormal fistula similar to that detected by CT. Ultrasonography also detected an accumulation of gas in both intrahepatic bile ducts, the common hepatic duct, and gallbladder, and gas bubbles in the biliary system moved with the patient's breathing. No signs of biliary atresia were noticed during the ultrasonographic examination.

Flexible fiberoptic bronchoscopy showed an abnormal opening at the right main bronchus; contrast was injected *via* the opening, and beside radiography was performed, which showed the outlines of the fistula, biliary tract, gallbladder, and

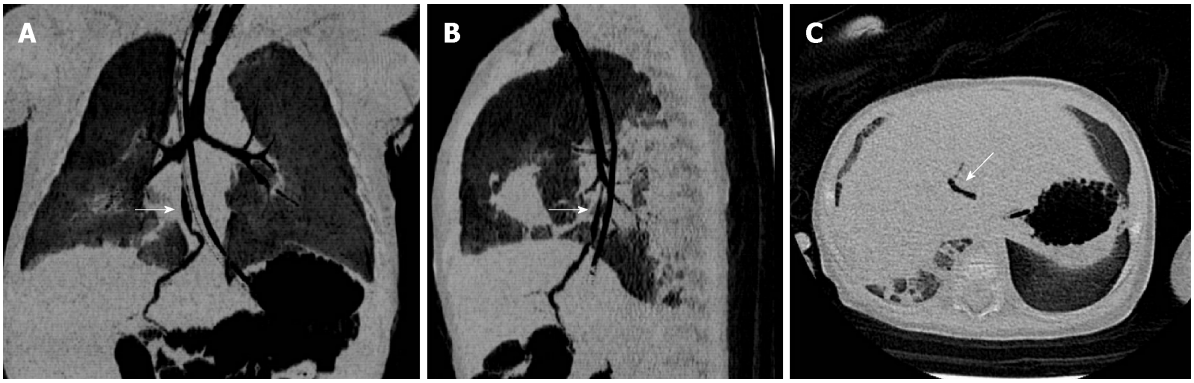

Figure 1 Computed tomography images. A and B: Computed tomography (CT) scan and tracheoesophageal reconstruction; the arrow indicates the abnormal connection between the right bronchus and biliary tract. C: CT scan; the white arrow indicates gas in the biliary tract.

duodenum (Figure 2).

FINAL DIAGNOSIS

Congenital bronchobiliary fistula.

TREATMENT

We initially performed laparoscopic biliary tract exploration and cholangiography to rule out biliary tract malformations. We found that the gallbladder was filled with green viscous bile, and the distal bile duct was patent. The abnormal fistula that bridged the intrahepatic bile duct and right bronchus was visible (Figure 3). Then, we performed posterolateral right thoracotomy at the sixth intercostal space. Green bile salt was deposited extensively beneath the visceral pleura. We dissected the mediastinal pleura and detected the fistula alongside the esophagus. The fistula formed down from the right bronchus to the right side of the hiatus, and it was about 4 mm in diameter and as soft as the esophagus. We punctured the lumen but produced nothing; then 10 mL of methylene blue was injected into the gallbladder, and the blue material was noticed subsequently at the puncture site and intratracheal tube (Figure 4). The fistula was confirmed accordingly. We separated the fistula and dissected it from the bronchus to the hiatus; both ends were securely ligated and sutured. Lastly, a thoracic drainage tube was placed.

OUTCOME AND FOLLOW-UP

On the day 3 postoperatively, the child was successfully weaned from the ventilator. She was in good condition with satisfactory pulmonary expansion and feeding tolerance. We attempted to remove the thoracic drainage tube but failed; subsequently, the baby developed pneumothorax and subcutaneous emphysema, which were considered due to improper operation, so the tube was reinserted. On day 7 postoperatively, the child developed cholestasis with a total bilirubin level of 71.6 μmol/L and conjugated bilirubin level of 58.1 μmol/L. Ultrasonography showed mild obstruction of the extrahepatic bile duct and slight dilation of the common bile duct. Two possibilities were considered: first, the extrahepatic bile duct was not patent enough to drain all the bile produced by the liver sufficiently; and second, inflammation and edema caused by the operation resulted in partial obstruction of the bile duct. Methylprednisolone was administered, and her bilirubin level decreased to normal range 3 d later. On day 10 postoperatively, growing chylous fluid was noticed in the drainage, so the oral feeding was withdrawn accordingly. Ten days later, the drainage gradually decreased, oral feeding was resumed without adverse reactions, and the drainage tube was removed. The baby recovered well and was discharged. She has been followed for 4 mo without any signs of discomfort.

A microscopic examination was performed separately for three parts of the removed fistula. Scattered mature and naive hyaline cartilage surrounded by clustered bronchial glands could be seen at the part near to the bronchus.

Figure 2 Contrast study via a fiberoptic bronchoscope. The contrast dye can be seen in the fistula, biliary tract, and duodenum, and the white arrow indicates the fistula.

Hyperplasia, angulation, aggregation, and columniation of the biliary glandular epithelial cells were found at the biliary end. The middle part was characterized by scattered, clustered glands, columniation of the glandular epithelia, squamous metaplasia, and cartilaginous tissue around the bronchial glands (Figure 5A-C).

DISCUSSION

CBBF can manifest as a series of symptoms caused by an abnormal connection between the hepatic duct and trachea or bronchus. The onset time and severity of symptoms are related to the diameter of the fistula; therefore, the symptoms can appear at any age from newborn to adulthood. However, most of them occur in newborns and infants. The main clinical features of CBBF are recurrent coughing, bilious sputum, or bile-stained sputum in tracheal intubation. Patients generally develop progressive dyspnea, cyanosis, severe pneumonia, respiratory distress syndrome, and even apnea; shortly after birth, mechanical ventilation is often needed. Bile-stained expectoration is the most typical sign among the aforementioned symptoms. It has been often misdiagnosed as an esophagotracheal fistula, gastroesophageal reflux, aspiration pneumonia, or high intestinal obstruction[4,5].

The first findings on CBBF were reported in 1952 by Neuhauser *et al*[6]. Altogether, there were 43 additional cases reported in English and 1 in Chinese thereafter (Table 1), and CBBF was more common in women than in men. An abnormal fistula can be opened at different positions around the carina: right main bronchus (20/44, 45.4%), carina (19/44, 43.2%), and left main bronchus (5/44, 11.4%). Fistulas generally passed through the esophageal hiatus and entered into the abdominal cavity; all fistulas communicated with the intrahepatic bile duct (left hepatic duct), except one fistula that opened at the common bile duct[7].

The pathogenesis of this disease is not clear yet. Abnormal development of the bronchial bud, which fuses with the bile duct, was regarded as the cause of the anomaly. Other reports postulated that CBBF was the result of digestive tract duplication and the upper respiratory tract[1,6,8-10].

As in most reports, the proximal part of the fistula had the characteristics of the respiratory tract, consisting of cartilage, pulmonary epithelia, glands, and smooth muscle, whereas the distal part was similar to the gastrointestinal tract. In our case, by examining the proximal, middle, and distal parts of the fistula, we found scattered mature hyaline cartilage, relatively naive cartilage, and clusters of bronchial glands at the bronchus end; whereas, glandular epithelia of the bile duct and interstitial cholestasis were found at the distal part of the fistula. In the middle part, the lumen was partly covered by pseudo-ciliated columnar epithelia. Cartilage rings lay beneath the mucosa. Co-existing single-layer columnar epithelium could also be seen in the same field. These histological findings could further verify the pathogenesis of CBBF.

CBBF was often complicated by biliary atresia, diaphragmatic hernia, esophagus atresia, and tracheoesophageal fistula, and the incidence of biliary malformation in this group was about 30%[11-15]. As long as this possibility was considered, the diagnosis of CBBF was not difficult. Magnetic resonance imaging, 3D-CT reconstruction, bronchoscopy, and ultrasonography can provide valuable clues.

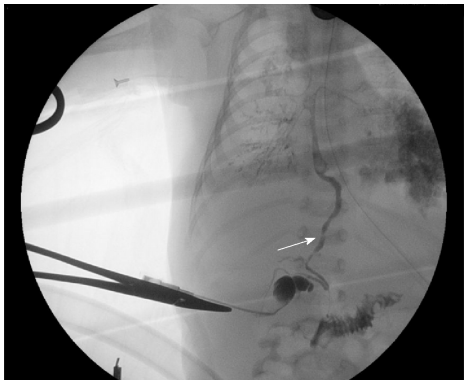

Figure 3 Intraoperative cholangiogram showing the connection between the biliary tract and right bronchus and patent distal bile duct; the white arrow indicates the biliary tract.

Bronchoscopy was the most common method for making a diagnosis, as it can be used to find the abnormal opening of the fistula and the overflowing bile via the opening[13,14,16-19]. 3D-CT reconstruction can accurately detect the existence of the fistula, which is of great significance for diagnosing CBBF. In the present case, the CT scan provided us with the first evidence of the abnormal fistula. Then fiberoptic bronchoscopy was performed, and a contrast dye was injected into the fistula once the opening was confirmed. Next, a radiographic examination was conducted; thus, the opening, shape, and connection of the fistula were all confirmed. The result of our treatment was excellent, and to our knowledge, this method has not been reported previously.

Surgical resection was the ultimate treatment of choice, but a specific method must be considered according to different anatomical structures[20-22]. According to previous reports, different kinds of operative methods have been used, such as hepatic lobectomy, anastomosis of the gallbladder or intestine with abnormal fistulas, and fistula resection. We think that it is necessary to consider whether there is a biliary tract anomaly when preparing an operative plan. If a biliary anomaly exists, biliary reconstruction must be considered[4,14]. We performed laparoscopic cholangiography first during the operation to exclude biliary atresia, and achieved a satisfactory result. We performed thoracotomy subsequently, and explored and identified the fistula in the mediastinum; the fistula formed down the right side of the esophagus. We punctured the lumen for assurance because there are many important structures nearby, and then we injected methylene blue into the gallbladder; the dye was seen flowing out of the puncture site, thus the fistula was confirmed. Because the extrahepatic biliary tract was normal, reconstruction was unnecessarily considered. Instead, we just separated the fistula and ligated and sutured it at the upper end and at the hiatus, and then the fistula was removed.

The overall mortality of CBBF was 15.91% (7/44) according to the literature, and three patients died of respiratory complications without surgery. Our patient experienced chylothorax shortly after thoracotomy, which may be due to injury of the thoracic duct when we dissected the fistula at the hiatus. Fortunately, she recovered after short-term conservative management. Cholestasis was another main complication encountered postoperatively, and ultrasonography showed slight dilatation of the bile duct. We evaluated her conditions and prescribed methylprednisolone. Cholestasis resolved after three doses; therefore, we deduced that the jaundice may have been due to temporary inflammation of the biliary tract.

CONCLUSION

Most surgeons may not encounter CBBF in their professional career considering its rarity. Its clinical symptoms are typical and special such as bile-stained expectoration. As long as surgeons have some insight into this kind of condition, its diagnosis and treatment are not difficult, and the prognosis would be excellent for most patients.

Table 1 All cases of congenital bronchobiliary fistula reported to date

N	Year	Author	PMID	Type of fistula	Age	Sex	Diagnostic method	Operative method	Complications	Therapeutic effects
1	2018	Netto *et al*[36]		Tracheal carina	21 d	F	CT	Surgical excision and ligation + selective left hepatectomy	None	Recovered after the hospital stay
2	2016	Pérez *et al*[7]	27606661	Right main bronchus	22 d	M	Bronchus examination and CT	Laparoscopic excision + left hepatectomy and cholangiopancreatostomy	Biliary dysplasia	Followed for 1 yr, normal
3	2016	Kwon *et al*[37]	27000583	Right main bronchus	17 yr	M	Bronchus examination and CT and MR imaging	Laparoscopic resection and ligation	None	Followed for 10 mo, normal
4	2015	Kumar *et al*[38]	26628766	Right main bronchus	3 yr	F	Bronchus examination	Surgical excision and ligation	None	Survived
5	2015	Kim *et al*[24]	25555959	Tracheal carina	17 yr	M	Bronchus examination and CT	Surgical excision and ligation	None	Survived
6	2015	Mo *et al*[25]	25617079	Right main bronchus	66 yr	M	Bronchus examination and CT	Wedge resection of the lung + resection of the intrahepatic tumor	Intrahepatic ductal papillary mucinous tumors	Survived
7	2015	Yu *et al*[26]	27307938	Tracheal carina	5 d	M	Bronchus examination and CT	Right thoracotomy + Kasia procedure	Biliary dysplasia	Followed for 6 mo, normal
8	2011	Hodgdon *et al*[27]	21827062	Right main bronchus	52 yr	F	CT	Surgical excision and ligation	None	Survived
9	2011	Sachdev *et al*[10]	21516501	Tracheal carina	2 yr 10 mo	M	Bronchus examination and CT	Surgical excision and ligation	None	Followed for 1 yr, normal
10	2011	Kumagai *et al*[28]	21297506	Tracheal carina	2 mo	F	Bronchus examination and MR imaging	Surgical excision and ligation	None	Followed for 7 mo, normal
11	2010	Tan *et al*[29]	20672712	Right main bronchus near carina	51 yr	F	Bronchus examination and CT	Surgical excision and ligation	None	Recovered after the hospital stay
12	2010	Croes *et al*[30]	20385268	Tracheal carina	3 d	F	Bronchus examination	Surgical excision and ligation	Biliary dysplasia	Survived
13	2009	Najdi *et al*[31]	19586763	Tracheal carina	6 d	F	Bronchus examination and CT	Surgical excision and ligation	Meconium aspiration syndrome	Died on day 2 after surgery
14	2009	Günlemez *et al*[22]	19101328	Left main bronchus	9 d	F	CT	Surgical excision and ligation	Extrahepatic biliary atresia	Followed for 9 mo, normal
15	2008	Uramoto *et al*[3]	18628589	Tracheal carina	65 yr	F	Bronchus examination and CT	No surgery	Lung cancer	Died of lung cancer within 28 mo since diagnosis
16	2008	Chawla *et al*[20]	18478221	Tracheal carina	< 28 d	M	CT	Surgical excision and ligation	Biliary dysplasia	followed for 6 mo, normal
17	2005	Aguilar *et al*[32]	16021209	Tracheal carina	6 yr	F	Bronchus examination	Surgical excision and ligation	None	Survived

18	2004	Hourigan *et al*[33]	14605784	Right main bronchus	13 d	M	MR imaging	Right thoracotomy + Kasia procedure	Biliary dysplasia	Survived
19	2002	DiFiore *et al*[15]	12149705	Right main bronchus	< 28 d	M	Intuition intraoperatively	Surgical removal + right diaphragmatic hernia repair	Right diaphragmatic hernia	Survived
20	2000	Duong *et al*[17]	10741248	Tracheal carina	3 yr	F	Bronchus examination	Surgical excision and ligation	None	Survived
21	2000	Tommasoni *et al*[14]	10922138	Tracheal carina	1 yr 9 mo	F	Bronchus examination	Nissen procedure + surgical excision and ligation	Gastroesophageal reflux	Followed for 6 yr, normal
22	2000	Tommasoni *et al*[14]	10922138	Tracheal carina	2 yr 6 mo	M	Bronchus examination and angiography	Surgical excision and ligation	Funnel chest	Followed for 4 yr, normal
23	1998	Fischer *et al*[34]	9854540	left main Bronchus	16 d	F	Bronchus examination	Surgical excision and ligation	None	Survived
24	1996	Egrari *et al*[35]	8783103	Tracheal carina	3 d	F	Bronchus examination	Surgical excision and ligation	None	Survived
25	1996	Nishimura *et al*[39]	8741536	Right main bronchus	57 yr	F	CT	Surgical excision and ligation	None	Died
26	1994	Ferkol *et al*[19]	8194299	Left main bronchus	23 d	M	Bronchus examination	Surgical excision and ligation	Biliary dysplasia	Died
27	1995	Tekant *et al*[5]	8035264	Tracheal carina	15 d	F	Bronchus examination	Surgical excision and ligation	Biliary dysplasia	Survived
28	1993	Gauderer *et al*[4]	8468661	Tracheal carina	2 yr	M	Bronchus examination	Surgical excision and ligation	Biliary dysplasia	Survived
29	1990	Yamaguchi *et al*[40]	2371608	Right main bronchus	32 yr	M	Bronchus examination	Surgical excision and ligation	None	Survived
30	1989	Mavunda *et al*[41]		Left main bronchus	1 yr	F	CT	Surgical excision and ligation	None	Survived
31	1988	de Carvalho *et al*[42]	3206388	Right main bronchus	32 yr	F	CT	Surgical excision and ligation	None	Followed for 1 yr, normal
32	1987	Levasseur *et al*[1]	3632120	Tracheal carina	22 yr	F	Bronchus examination	Surgical excision and ligation	None	Survived
33	1986	Lindahl *et al*[18]	3746610	Right main bronchus	15 d	F	Bronchus examination	Surgical excision and ligation	None	Survived
34	1985	Chang *et al*[9]	4001424	Right main bronchus	12 h	M	Bronchus examination	Surgical excision and ligation	None	Survived
35	1984	Chan *et al*[12]	6697133	Right main bronchus	4 d	F	Not mentioned	Surgical excision and ligation	Biliary dysplasia	Died
36	1976	Kalayoğlu *et al*[11]	957073	Right main bronchus	4 d	F	Not mentioned	Surgical excision and ligation	None	Died
37	1974	Cuadros *et al*[43]		Tracheal carina	6 yr	M	Bronchus examination	Surgical excision and ligation	None	Survived
38	1971	Sane *et al*[8]	5573334	Right main bronchus	4 wk	F	Bronchus examination	Surgical excision and ligation	None	Survived

39	1970	Wagget *et al*[44]		Left main bronchus	3 wk	F	Bronchus examination	Surgical excision and ligation	Biliary dysplasia	Survived
40	1968	Weitzman *et al*[45]	5667415	Right main bronchus	2 yr 9 mo	M	Bronchus examination	Surgical excision and ligation	None	Survived
41	1966	Stigol *et al*[2]	5902432	Right main bronchus	1 yr 2 mo	F	Bronchus examination	Surgical excision and ligation	None	Survived
42	1963	Enjoji *et al*[46]		Right main bronchus	7 mo	M	Biopsy	No surgery	Biliary dysplasia	Died
43	1952	Neuhauser *et al*[6]	14914166	Right main bronchus	5 mo	F	Bronchus examination	No surgery	None	Died
44	2013	Zheng *et al*[23]		Tracheal carina	27 d	F	Bronchus examination and CT	Surgical excision and ligation	Biliary atresia	Survived

CBBF: Congenital bronchobiliary fistula; CT: Computed tomography; MR: Magnetic resonance; F: Female; M: Male.

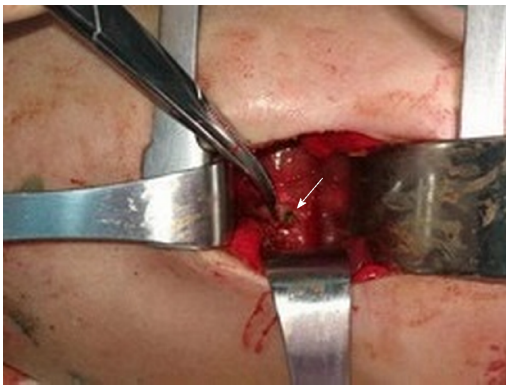

Figure 4 Methylene blue was injected into the gallbladder, and it flowed out from the puncture site; the white arrow indicates the puncture site.

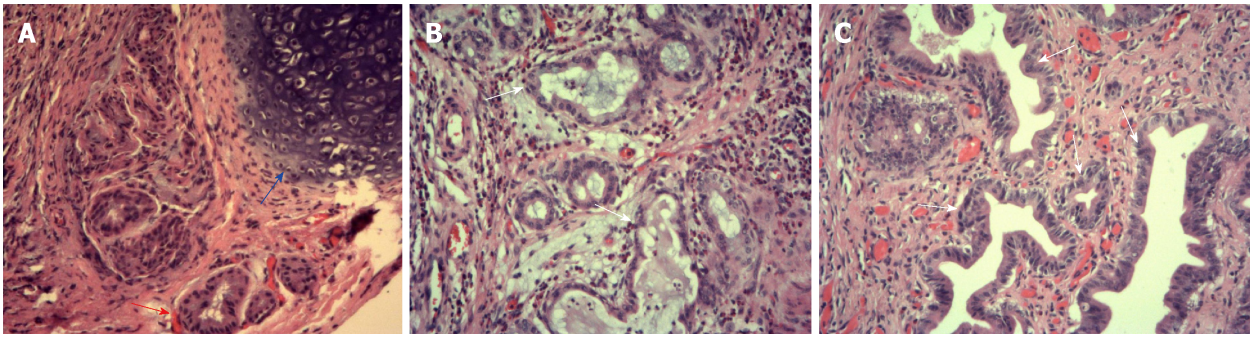

Figure 5 A microscopic examination performed separately for three parts of the removed fistula. A: The red arrow indicates the lumen that was partly covered by pseudo-ciliated columnar epithelium. The blue arrow indicates the cartilage rings that lay beneath the mucosa; B: The arrow indicates the clusters of bronchial glands at the end of the bronchus; C: The arrow indicates the glandular epithelia of the bile duct and interstitial cholestasis.

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