

**Case Report**

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**Diagnosis of congenital bronchobiliary fistula in children by bilirubin crystallization in the bronchoalveolar lavage fluid: A case report and literature review**

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**Abstract**

**Background:** Congenital Bronchobiliary Fistula (CBBF) is a rare disease with abnormal connection between the biliary tract and the bronchus. The misdiagnosis rate of CBBF is high, delayed surgery may induce poor outcome.

**Case presentation:** We reported a girl with CBBF who was confirmed via bronchoscopy and chest Computed Tomography (CT). The girl was 7-month-old admitted to the hospital with "recurrent pneumonia". Bilirubin crystallization detection was positive in the Broncho Alveolar Lavage Fluid (BALF). CT images showed the abnormal traffic between the biliary tract and the right main bronchus, the girl was diagnosed with CBBF and recovered after surgical operation.

**Literature search:** A total of 27 cases were collected, 14 (51.8%) cases were diagnosed in the neonatal period, 9 (33.3%) patients were diagnosed in infancy. CBBF was more common in female (19/27, 70.3%). The specific manifestations were bilious sputum (24/27, 88.9%) and bilious vomiting (7/27, 22.2%). Recurrent pneumonia (9/27, 33.3%) was common clinical manifestations. The most abnormal fistula originates different location around the carina was right main bronchus (21/27, 77.8%). There were 23(85.2%) cases recovered after surgical operation and 4(14.8%) cases with severe comorbidities/complications died.

**Conclusions:** CBBF should be suspected in infants with bilious sputum. Bronchoscopy plays crucial role in preoperative evaluation and postoperative follow-up. With early diagnosis and surgery, the prognosis is quite well for CBBF patients without severe biliary malformation and/or severe postoperative complications.

**Keywords:** Congenital Bronchobiliary Fistula (CBBF); bronchoalveolar lavage fluid; bilirubin crystallization; children; case report.

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## Background

Congenital Bronchobiliary Fistula (CBBF) refers to the abnormal traffic between the biliary system and the bronchial tree. CBBF is a rare disease and easily misdiagnosed as pneumonia or other diseases in clinical practice, which resulting in delayed treatment and worse outcomes [1,2]. Few studies have reported the role of bilirubin crystallization detection in the Broncho Alveolar Lavage fluid (BALF) for diagnosis CBBF [3]. Here, we reported an infant with recurrent pneumonia who admitted to Children`s hospital, Chongqing Medical University. She was diagnosed with CBBF combined positive bilirubin crystallization detection in BALF with related chest CT images and recovered after surgery. The detection of bilirubin crystallization in BALF plays important diagnostic value in patients suspected with CBBF [3,4]. Surgery is the first choice for CBBF patients [3-6]. Next, we collected literature about patients with CBBF so as to further improve the recognition and early intervention.

## Case presentation clinical data

A 7-month-old girl was admitted to respiratory department with persistent wet cough and recurrent wheezing. She was diagnosed with recurrent pneumonia and treated with antibiotics, bronchodilators and glucocorticoids empirically when symptoms worsen. She was full-term with normal manifestation during perinatal period. She occasionally expectorated bright-yellow sputum when drinking breast or milk (Figure 1). Chest Computed Tomography (CT) images showed an abnormal bronchial bifurcation originated from the right main bronchus (Figure 2A). Both transverse (Figure 2B) and anamorphic (Figure 2C) chest CT indicated gas shadow anterior to the esophagus at the diaphragm level. Yellow, serous secretions emerging from an anomalous orifice located at the right main bronchus via bronchoscopy (Figure 3). The bright-yellow BALF was obtained and tested for bilirubin with positive detection. The contrast agent meglumine was injected through the anomalous orifice via flexible bronchoscope. Chest CT examination showed the contrast agent entered the abnormal bronchus and traveled downward, passing through the left hepatic duct, the common hepatic duct and common bile duct (Figure 4). The girl was diagnosed with Congenital Broncho Biliary Fistula (CBBF) based on these findings. She had operation to remove the anomalous fistula. During the ten months following-up, the girl was absent from wet cough, wheezing and pneumonia. Chest computed tomography examination demonstrated that the anomalous orifice and lung shadow disappeared (Figure 2D). Bronchoscopy showed an anomalous diverticulum (Figure 3C) and the BALF was colorless with no bilirubin presented. Pathological examination showed tubular structure of cartilage and muscle, lined with stratified squamous epithelium, pseudostratified columnar ciliated epithelium and submucosal glands (Figure 5).

## Literature review

### Information sources and search key words

Using the search terms “congenital bronchobiliary fistula” OR “bronchobiliary fistula”, Medline (via PubMed), Embase and web of science databases, China National Knowledge Infrastructure (CNKI) and Wan fang were searched. We also searched Chinese Medical Care Repository, searches for relevant articles



Figure 1: Bright-yellow sputum.

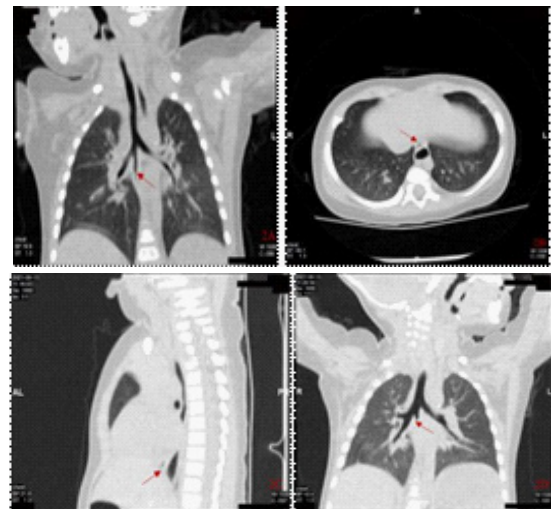


Figure 2: A: Airway reconstruction showed a tubular shadow at the terminal of the right main bronchus before surgery (red arrow); B: Transverse view showed a gas shadow in front of the esophagus at the level of the diaphragm (red arrow); C: Loss of shape showed the diaphragm with gas shadow in front of the esophagus (red arrow); D: Airway reconstruction showed tubular shadow disappeared with a blind end after surgery (red arrow)

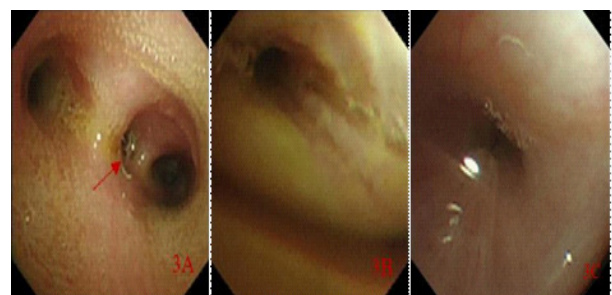


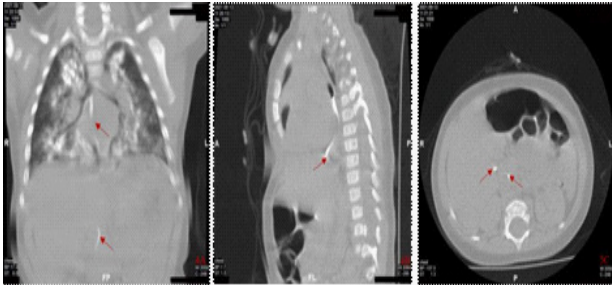
Figure 3: A: An anomalous orifice was found at the right main bronchus under bronchoscopy (red arrow); B: Yellow secretions from the anomalous orifice (before surgery); C: an anomalous diverticulum after surgery.

were performed with the following items: “congenital bronchobiliary fistula” OR “bronchobiliary fistula. Searches were limited to articles published in English till March 31, 2022.

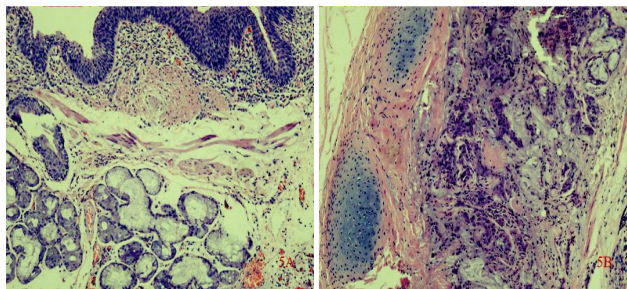
## Eligible criteria

### Inclusion

Congenital bronchobiliary fistula patients, not restricted by age, gender, disease course, race, region and other factors;



**Figure 4:** A: Airway reconstruction showed that the contrast agent entered the abnormal bronchus and traveled downward (red arrows); B: The contrast agent entered the abnormal bronchus and traveled down through the diaphragm (red arrow). C: Cross-sectional view showed the inflow of contrast agent into the common bile duct and common hepatic duct (red arrows).



**Figure 5:** Pathological examination of the anomalous fistula A: tubular structure of cartilage and muscle, lined with stratified squamous epithelium, pseudostratified columnar ciliated epithelium and submucosal glands, inflammatory infiltration of lymphocytes in the mucosal lamina propria; B: glandular hyperplasia, dominated by mucus glands. Mucus exuded into the periductal interstitial space.

#### Exclusion:

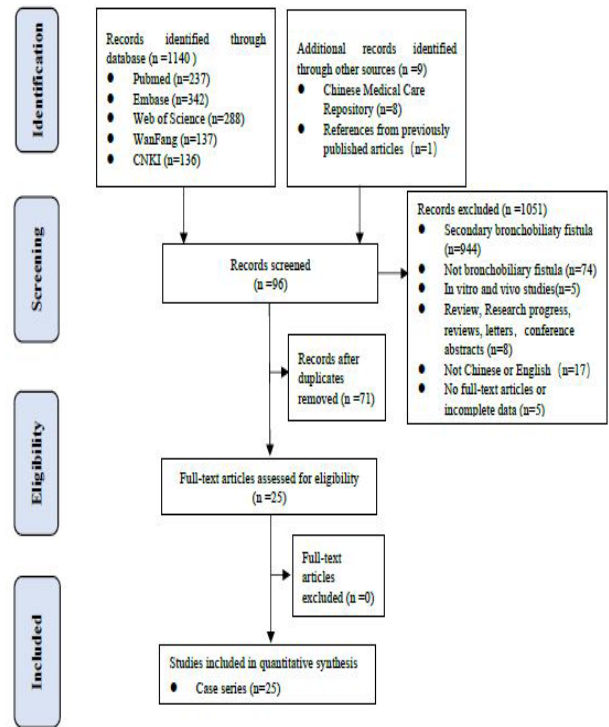
- 1) Secondary bronchobiliary fistula patients;
- 2) Article not published in Chinese or English;
- 3) In vitro and in vivo studies;
- 4) Conference abstracts;
- 5) Full-text or data cannot be extracted Searching results

Searches of literatures initially identified 1149 potentially relevant records. Following review of the title and abstract, 1051 records were excluded, and a further 73 duplicated records were excluded, 25 records were included. Therefore, 19 English articles and 6 Chinese articles were included, a total of 26 cases [4-28]. Combined with 1 case in this article, a total of 27 cases of CBBF were enrolled. The clinical data summary table is shown in Table 1.

#### Discussion

CBBF is a rare disease with congenital abnormal connection between the hepatic duct and trachea or bronchus. The age of onset and severity of symptoms depend on the diameter of the fistula.

Therefore, the symptoms appear at any age from newborn to adults [9,10,29]. Typical clinical feature of CBBF is bilious sputum or bile-stained sputum in tracheal intubation. Patients usually have chronic wet coughing, recurrent wheezing, shortness of breath and growth retardation [12]. CBBF has usually been misdiagnosed as an esophagotracheal fistula, gastroesophageal



**Figure 6:** Flow diagram of congenital bronchobiliary fistula.

reflux, aspiration pneumonia, tracheoesophageal fistula or high intestinal obstruction [30,31].

In 1952, Neuhauser et al [29] reported the first case of CBBF. Up to now, 27 cases have been enrolled in English and Chinese (Table 1). Among them, fourteen (51.8%) cases were diagnosed in the neonatal period, nine (33.3%) were diagnosed in infancy, four (14.8%) were diagnosed in puberty or adulthood. The common manifestations were bilious sputum (24/27, 88.9%), recurrent pneumonia (9/27, 33.3%) and bilious vomiting (7/27, 22.2%). CBBF was more common in female (19/27, 70.3%). The abnormal fistula originates different location around the carina: right main bronchus (21/27, 77.8%), left main bronchus (3/27, 11.1%), right intermediate bronchus (2/27, 7.4%), and bilateral main bronchus (1/27, 3.7%).

The mechanism of CBBF formation is not clearly clarified. However, there are two possible speculated mechanisms. One is considered as the duplication of the upper gastrointestinal tract, which growing between the laryngotrachea and hepatic diverticulum; the other is believed to be the fusion between abnormal bronchial buds and abnormal bile ducts [20,26]. The pathological results in our case consistent with the latter pathogenesis.

The detection of bilirubin crystallization in sputum or/and BALF plays crucial role in CBBF diagnosis [4,5,11-14,29,31]. Furthermore, abnormal fistula was easily found via bronchoscopy. In this case, the bilirubin detection was positive in the BALF and the location of the fistula connection was confirmed via bronchoscopy and fistula angiography, which provided more comprehensive information and was verified intraoperatively.

Chest CT and airway reconstruction, MRI or isotope examination can find an abnormal fistula from the trachea, through the diaphragm, into the abdominal cavity, and liver lobes communicate with each other [15]. Intraoperative management of fistula could base on preoperative isotope examination [32]. Liu AH et al [31] indicated gas accumulation in the lumen of the



**Table 1:** All cases of congenital bronchobiliary fistula reported to data.

N	Year	Author	Type of fistula	Age	Sex	Diagnositc method	Comorbidity	Operative method	Therapeutic effects
1	2021	Meng EY et al	Right main bronchus	3d	F	Bronchoscopy and CT	biliary atresia	Surgical excision and ligation + Roux En-Y anastomosis	Survived
2	2021	Wang Q et al	Right intermediate bronchus	7 yr 6 mo	F	Bronchoscopy and MRI	None	Surgical excision and ligation	Survived
3	2019	Wu XM et al	Right intermediate bronchus	3 yr 5 mo	F	Bronchoscopy and CT and SPECT	None	Surgical excision and ligation	Survived
4	2018	Chen X et al	Right main bronchus	3 yr 7 mo	F	Bronchoscopy and MRCP	None	Bioadhesive occlusion	Followed for 4 yr 3 mo, normal
5	2013	Li K et al	Right main bronchus	27 d	F	Bronchoscopy and CT	Extrahepatic biliary atresia	Surgical excision and ligation +Fistula distal-jejunal Roux En-Y anastomosis	Followed for 3 mo, normal
6	2010	Ge Y et al	Right main bronchus	51 yr	F	CT	None	Surgical excision and ligation	Survived
7	2021	Bing Z et al	Right main bronchus	2 yr	M	Bronchoscopy and CT	None	Surgical excision and ligation	Followed for 2 yr 6 mo, normal
8	2021	Thuong Vu et al	Right main bronchus	2 m	F	Bronchoscopy and CT	None	Surgical excision and ligation	Survived
9	2019	Li TY et al	Right main bronchus	5 d	F	Bronchoscopy and CT and ultrasound	None	Surgical excision and ligation	Followed for 4 mo, normal
10	2016	NaKJ et al	Right main bronchus	17 yr	M	Bronchus examination and CT and MR imaging	None	Laparoscopic resection and ligation	Followed for 10 mo, normal
11	2009	Günlemez et al	Left main bronchus	9 d	F	CT	Extrahepatic biliary atresia	Surgical excision and ligation	Followed for 9 mo, Normal
12	2004	Hourigan et al	Right main bronchus	13 d	M	MRI	Biliary Dysplasia	Right thoracotomy + Kasia procedure	Survived
13	2002	DiFiore et al	Right main bronchus	<28 d	M	Intuition intraoperatively	Right diaphragmatic hernia	Surgical removal +right diaphragmatic hernia repair	Survived
14	1994	Ferkol et al	Left main bronchus	23 d	M	Bronchus examination	Biliary dysplasia	Surgical excision and ligation	Died
15	1993	Gauderer et al	Bilateral main bronchus	3 w	F	Bronchoscopy and angiography	None	3 times thoracotomy (fistula clipping et al) + the hepatic left loberection	Followed for 8 yr, survived
			Left main bronchus	1 y	F	Bronchoscopy and hepatobiliary scintigraphy	None	Surgical excision and ligation and Roux En-Y anastomosis	Followed for 6y, normal
16	1990	Yamaguchi et al	Right main bronchus	32 yr	M	Bronchus examination	None	Surgical excision and ligation	Survived
17	1988	de Carvalho et al	Right main bronchus	32 yr	F	CT	None	Surgical excision and ligation	Followed for 1 yr, normal
18	1986	Lindahl et al	Right main Bronchus	15 d	F	Bronchus examination	None	Surgical excision and ligation	Survived
19	1985	Chang et al		12 h	M	Bronchus examination	None	Surgical excision and ligation	Survived
20	1984	Chan et al	Right main bronchus	4 d	F	Not mentioned	Biliary Dysplasia	Surgical excision and ligation	Died
21	1976	Kalayoglu et al	Right main bronchus	4 d	F	Surgical findings	Esophageal atresia Tracheoesophageal fistula	Surgical excision and ligation	Died
22	1971	Sane et al	Right main bronchus	4 wk	F	Bronchus examination	None	Surgical excision and ligation	Survived
23	1970	Wagget et al	Left main Bronchus	3 wk	F	Bronchus examination	None	Surgical excision and ligation	Survived
24	1968	Weitzman et al	Right main Bronchus	2 yr 9 mo	M	Bronchus examination	None	Surgical excision and ligation	Survived
25	1952	Neuhauser et al	Right main bronchus	5 mo	F	Bronchus Examination and CT	None	No surgery	Died

right hepatic duct and the gallbladder lumen by hepatobiliary B-ultrasound, which provided clues for clinical diagnosis of BBF.

Patients with CBBF are ineffective to routine treatment and require operation to remove the fistula. If the bile drainage is normal, only thoracic fistula resection can be performed [33,34]; there also has reports of resection of thoracic and abdominal fistulas [13]. Fistula-jejunostomy Roux-en-Y anastomosis, hilar-jejunostomy, or Cholecystojejunostomy can be performed to fully drain bile so as to avoid fistula recurrence for cases with biliary malformation, such as absence of common bile duct, abnormal bile-intestinal drainage [14].

As surgery has higher cost and more damage, interventional therapy is alternative for CBBF patients without severe biliary malformation. The bronchial occlusion with biological glue has achieved good results in the treatment of adult respiratory diseases [35,36]. Tissue glue occlusion via bronchoscopy also have been used in the treatment of adult secondary bronchobiliary fistulas [36-38]. Chen X et al [7] successfully cured the pediatric patient with congenital bronchobiliary fistula for the first time via bronchoscopy. The interventional treatment via bronchoscopy in patients with bronchobiliary fistula requires respiratory intervention doctors to achieve further breakthroughs in future.

Combined this case and literature review, twenty-three (85.2%) patients recovered after operation, four (14.8%) patients died from severe comorbidities (biliary dysplasia (n=1), biliary atresia (n=1), congenital diaphragmatic hernia (n=1), esophageal atresia and/or tracheoesophageal fistula (n=1)). Two cases had complications (pneumothorax (n=1), septicemia (n=1)). One case with no surgery. CBBF patients with delayed or no operation, severe biliary malformation, or severe postoperative complications have poor prognosis [17,23,24,28].

## Conclusion

In conclusion, CBBF should be suspected in infants with biliary sputum. Besides, bronchoscopy plays crucial role in pre-operative evaluation and postoperative follow-up. With early diagnosis and surgery, the prognosis is good for CBBF patients without severe biliary malformation and/or severe postoperative complications.

## Declarations

**Authors' contributions:** Conception and design: GL Z, ZX L; Administrative support: ZX L, J L; Bronchoscopy and bronchoscopic figures: CJ W, GL Z; Provision of study materials or patients: ZL W, XY T, YY L, QY L, XJ X; Imaging and pathological figures: H D, J Z; Manuscript writing: GL Z, ZX L; Final approval of manuscript: All authors.

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**Availability of data and materials:** Not applicable.

**Declarations Ethics:** All procedures performed in studies involving human participant was in accordance with the ethical standards of the Children's Hospital of Chongqing Medical University research committee. Consent for publication.

**Consent form:** An informed consent was signed by the patient's mother who agreed to publish this case.

**Competing interests:** No competing interests for all authors.

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