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Case Report

Bilioptysis Secondary to Broncho-Biliary Fistula- A Rare Complication of Liver Abscess in a Pediatric Patient: A Comprehensive Case Report

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ABSTRACT

Broncho-biliary fistula (BBF), causing bilioptysis and persistent pneumonia is a rare complication of liver abscess in pediatric population. Only one case has been reported in pediatric literature. We report a case of a 4-year-old child, treated outside for liver abscess then presented to us with persistent bilious expectoration and respiratory distress. Computed tomography revealed a focal rent in the diaphragm with communication with segmental bronchi of right lower lobe. The child underwent thoracoscopic dissection of right lower lobe and repair of the rent. The child was asymptomatic and clinically well on follow-up after 1 month.

INTRODUCTION

Although liver abscess is a prevalent disease among the pediatric population worldwide, broncho-biliary fistula is known as a very rare complication [1,2]. Broncho-biliary fistula (BBF) consists of an abnormal communication between the biliary tract and the bronchial tree. It can present with chronic cough, persistent pneumonia, and bilioptysis i.e. expectoration of bile in the sputum [3]. Reported cases are either congenital in etiology or acquired post-abdominal procedures, infections, trauma, or tumors, particularly in adult patients [3]. Only a few cases of acquired BBF are reported in pediatric patients and almost all of them were either acquired traumatically or secondary to hydatid cysts or malignancies. Hence, the occurrence of BBF as a complication of a liver abscess, along with the presentation of bilioptysis, is exceptionally rare, especially within the pediatric population. We report a case of acquired BBF in a 4-year-old child developed as a complication of liver abscess.

CLINICAL DESCRIPTION

A 4-year-old girl child presented to the Department of Pediatrics of a tertiary care centre in New Delhi, India, with complaints of high-grade fever, pain abdomen, cough, and vomiting, she was then evaluated and on Ultrasonography (USG) a 42cc liver abscess was present and on chest x-ray right lower zone consolidation was present. The child was then treated for liver abscess with right lower zone pneumonia conservatively, with IV antibiotics for 3 weeks, and was discharged on oral antibiotics for 2 weeks. The child then remained mostly well at home except for a persistent cough which was productive with greenish-tinged yellow sputum, (Figure 1) not responding to anti-tussives and mucolytics she took by herself at home.

Figure 1: Bilioptysis: yellowish-green coloured bilious sputum.

She again presented to the pediatric emergency of our hospital, 2 months after discharge with complaints of high-grade fever for the last 4 days associated with persistent productive cough, and fast breathing for the last 2 days. On presentation, she was febrile with significantly increased work of breathing. She had tachycardia with a heart rate of 118/min and a respiratory rate of 46/min with Subcostal and Intercoastal Retractions. Peripheral pulses were well palpable with oxygen saturation of 86% under room air. On respiratory system examination, movements of the right hemithorax with respiration were reduced, with bronchial breath sounds in the right inframammary, infra-axillary, and infrascapular areas with crackles on auscultation. Per abdomen, examination revealed a palpable liver 1 cm below the costal margin, soft and non-tender with a span of 7 cm. The spleen was not palpable. Central nervous system and cardiovascular system examinations were within normal limits.

MANAGEMENT AND OUTCOME

On investigating, the hemogram revealed hemoglobin of 11.3g/dL with total leucocyte count of 5300/mm3 with 53%polymorphs and 39% lymphocytes, and a platelet count of 4.02Lac. Her liver function test revealed Total Bilirubin of 0.4 mg/dL with Aspartate aminotransferase/ Alanine Aminotransferase of 31/17 IU/L respectively and ALP of 142 U/L with total proteins and serum albumin values of 7.7g/dL and4.1 g/dL respectively. Kidney function tests were within normal limits with serum urea and creatinine levels of 17mg/dL and 0.2mg/dL respectively. Gross examination of sputum revealed thick greenish-yellow sputum, with positive bilirubin levels on dipstick-examination. Bilirubin levels of sputum were planned but could not be done to restrained resources.

Chest X-ray revealed a right lower zone non-homogenous patch with air bronchogram with blunting of the right costophrenic angle (Figure 2). USG abdomen revealed a normal liver with residual liver abscess of size approximately10 cc, and USG chest revealed consolidation in the right lower zone with minimal pleural fluid and the left costophrenic angle was clear. Workup for tubercular aetiology was done, the Mantoux test was 3mm(negative), and Sputum and Gastric Aspirates for AFB and CBNAAT were negative.

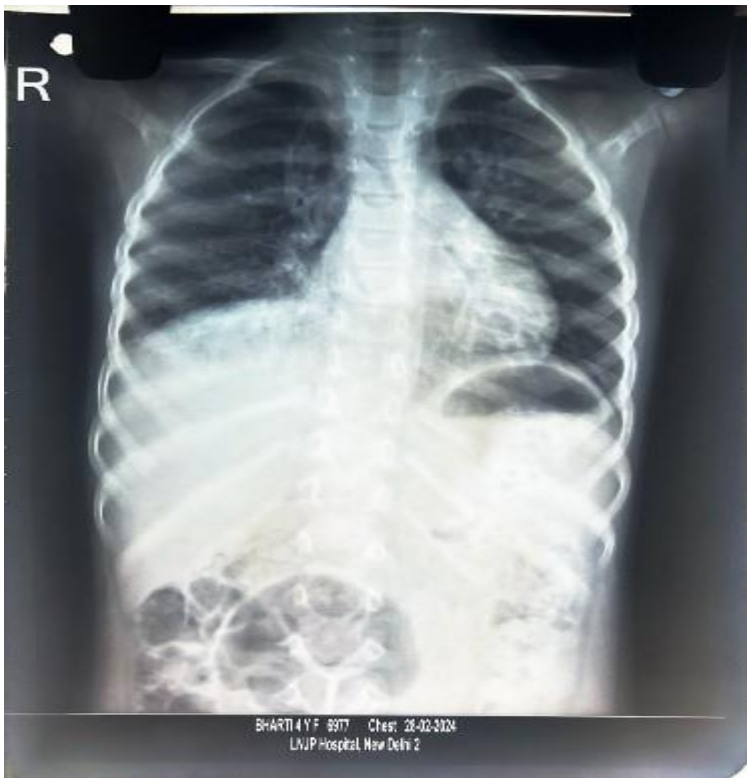

Figure 2: Chest X-ray posteroanterior view of the child (a) On first admission showing right lower zone non-homogenous opacity with air-bronchogram and obliterated right costophrenic angle and (b) before surgical procedure showing persistent of consolidation.

The child was kept on oxygen support via nasal prongs, was started on appropriate antibiotics according to hospital protocol. A workup for recurrent pneumonia was planned. 2D-ECHO of the heart revealed a normal study. Workup for immunodeficiency disorders showed negative HIV serology, normal Immunoglobulins and

complement levels, normal CD4 and CD8 counts, and a normal Nitro blue tetrazolium dye test for Chronic Granulomatous disease.

In view of persistent bilioptysis, CECT thorax was done which revealed a focal rent in the right hemidiaphragm at its posterior aspect with communication with lateral basal segmental bronchi of right lower lobe. With this finding and suspicion of broncho-biliary fistula the patient was taken up for thoracoscopic intervention. On thoracoscopy, after dissection of right lower lobe, which was densely adherent with the diaphragm, a focal rent of 2x2 cm in the diaphragm (Figure 3) was visible with superior surface of liver clearly visible through the defect. The rent was repaired thoracoscopically with nonabsorbable sutures, and no other rent could be identified in the diaphragm.

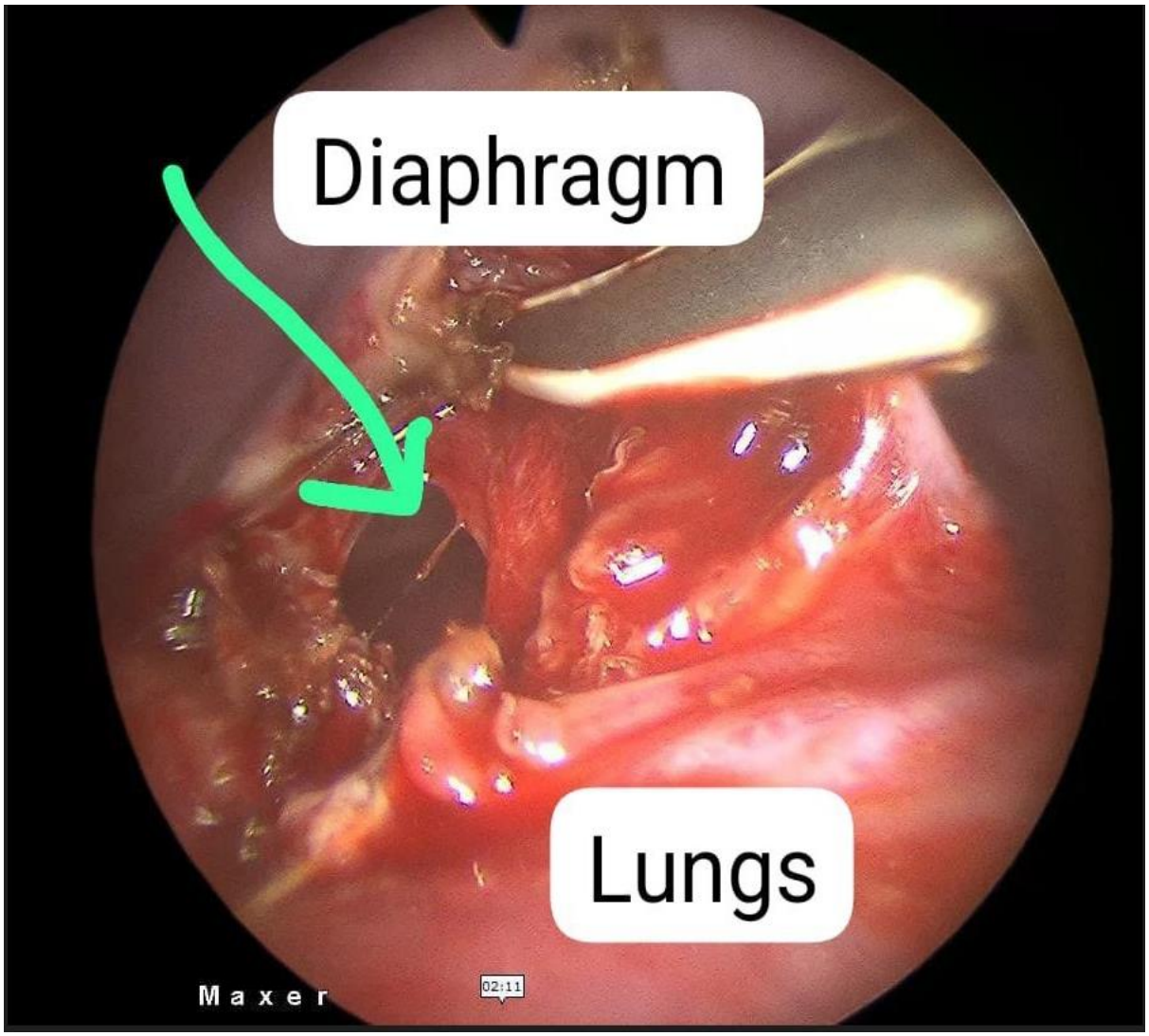

Figure 3: Thoracoscopic image showing an opening in the diaphragm (arrow) with diaphragm (superiorly) and lung tissue (inferiorly).

Postoperatively, patient was kept under observation with 2 intercoastal drainage(ICD) tubes insitu. After normal recovery of the patient the ICDs were removed and the patient was discharged on room air without any respiratory

difficulty. On follow-up after 1 month, child was asymptomatic, the thoracotomy scar healed (Figure 4), she gained weight and was thriving well.

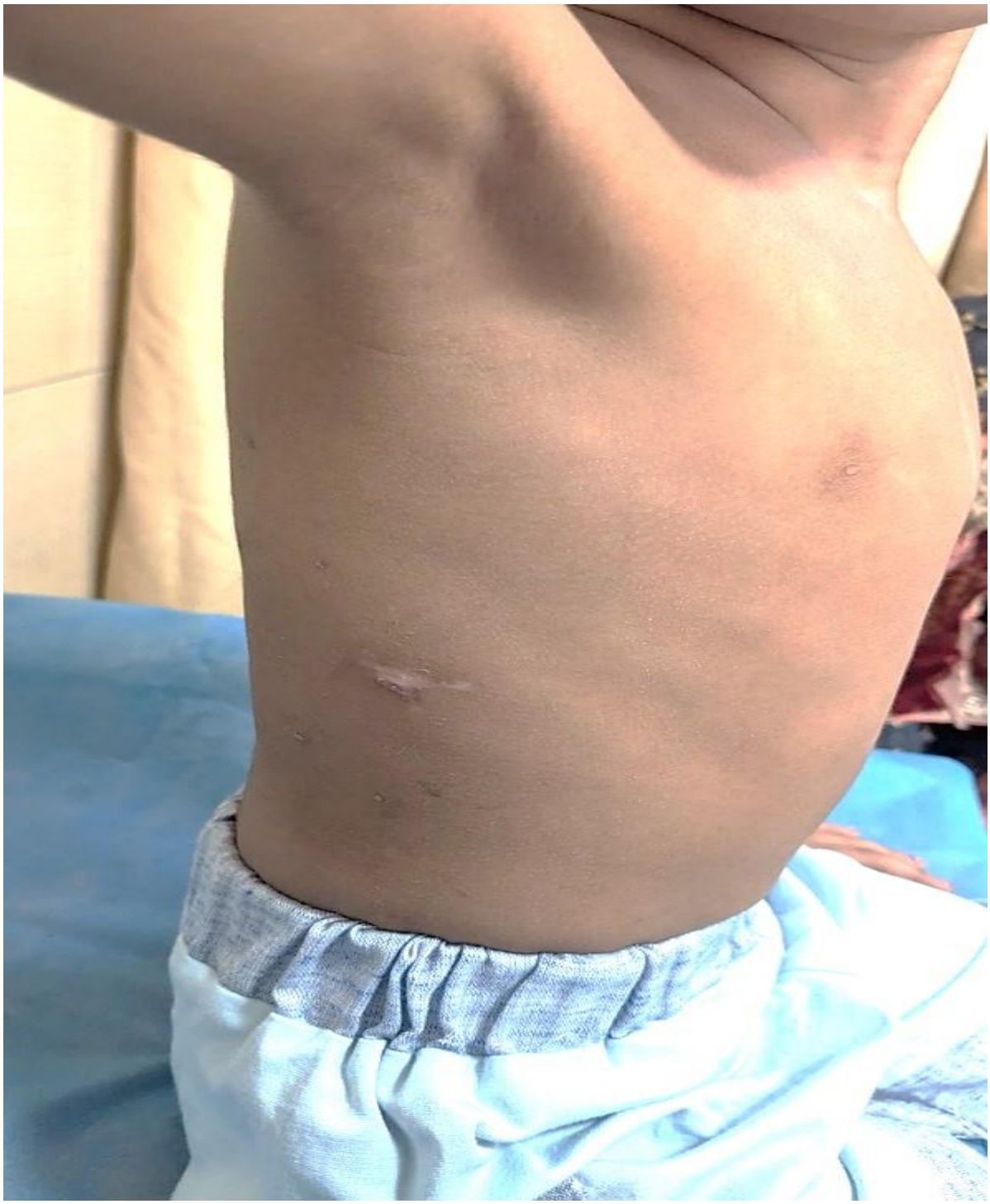

Figure 4: Lateral view of chest of the child, post discharge showing healed thoracoscopic scar.

DISCUSSION AND REVIEW OF LITERATURE

Broncho-biliary fistula (BBF) in children is an uncommon yet clinically significant entity characterized by an aberrant connection between the biliary tree and the bronchial system. The incidence of broncho biliary fistula in adults is reported to be around 2.5-16% [9] however, with only a few reported cases in the literature, its incidence

and precise epidemiological data in the pediatric population arimited. The etiology of BBF in children can be broadly classified into congenital and acquired types, with the latter developing as a complication of subphrenic liver abscesses, hydatid cysts, malignancies, abdominal procedures, and trauma [1,4].

We reported a case of an acquired broncho biliary fistula caused by a ruptured liver abscess in a pediatric patient. From our PubMed search and review of literature on acquired broncho biliary fistula in pediatric patients, only one case report of BBF as a complication of liver abscess is reported [1]. Table 1 provides information on the previously published cases of acquired BBF in children reported worldwide.

Table 1: Review of literature on Acquired Broncho biliary fistula in children.

Author, place, year	Age at diagnosis, gender	Primary diagnosis	Interval between primary diagnosis and BBF (months)	Clinical features	Imaging/ diagnostic modality	Treatment	Outcome as per last follow-up
Kumar et al., India, 2015.[1]	3.5 years, female	Ruptured liver abscess	7	Biliopt ysis	MRCP	Right lateral thoracotmy and right lower lobectomy with surgical excision of sinus tract	Alive and healthy
Yahya et al, Morocco, 2023. [2]	7 years, male	Liver hydatid cyst	6	Biliopt ysis	CT chest and abdomen	Oral albendazole therapy	Unknown
Rahimi et al.,Afganistan, 2023. [4]	17 years, female	Liver hydatid cyst	10	Biliopt ysis	CT chest and abdomen	Right thoracotomy. Adhesiolysis, and fistula tract excison	Alive and healthy
Yang et al., China,2017. [5]	10 years, male	Traumatic liver rupture	36	Biliopt ysis	CT could no identify the fistula	Orthotopic liver transplantation (persistent BBF and HVOTO)	Alive and healthy
Gautam et al, India, 2023. [6]	7 years female	Wilson disease with live donor liver transplant	3	Biliopt ysis	Bronchosc opy	Laparotomy, fistula tract excision, Roux-en-Y hepaticojejeunostomy and subphrenic drain	Alive and healthy
Copraciogl u F et al, Turkey, 2006. [7]	12 years, male	Undifferentiated sarcoma of liver	6	Biliopt ysis	Hepatobili ary scintigraph y	Chemotherapy and right thoracotomy and excision of fistulous tract	Unknown
Zainal et al., Malaysia, 2020. [8]	1 year, male	Hepatoblastoma	6	Biliopt ysis	Intraoperat ively	Right thoracotomy and lower lobectomy	Alive and healthy

The pathological connection in BBF leads to the passage of bile into the respiratory tract. The caustic nature of the bile can induce a chemical erosion of the diaphragm, lung, and pleura, preventing its spontaneous closure and resulting in a spectrum of respiratory and hepatobiliary symptoms. Biliopystsis, i.e. expectoration of yellowish

bilious sputum, as seen in our case, is pathognomonic of BBF reported in most of the case reports. Fever, persistent productive cough with copious sputum, fast breathing, and dyspnea which were present in our case were other common features reported [1-8]. Other features, such as jaundice, abdominal pain, and vomiting, may be seen which could be explained by the obstruction to the flow of bile or underlying primary disease [1-8].

The diagnosis relies on confirming the anomalous tract. The interval between the primary diagnosis and the detection of BBF ranged between 3 to 36 months [1,2,4-8]. The rarity of this complication and the inconclusive routine investigations lead to a lag in the diagnosis. Gross and biochemistry evaluation may provide evidence of bile in the sputum [5]. Ultrasonography, which is commonly used for primary diagnosis and therapeutic interventions in liver abscess, the visualization of the tract is very difficult and is often non-conclusive in providing the diagnosis of BBF. CT chest and abdomen is the preferred and most used imaging modality with findings delineating the location and associated pulmonary and bronchial Figure 2 and 3 Magnetic resonance cholangiopancreatography (MRCP)1 and hepatic scintigraphy[7] are other non-invasive diagnostic modalities where as bronchoscopy [6]and endoscopic retrograde cholangiopancreatography (ERCP) are the invasive methods with additional therapeutic possibilities. Intraoperatively, fistula has also been demonstrated by cholangiography [8].

Treatment of the primary disease, as provided in our case is with adequate antibiotics, chest physiotherapy to provide adequate clearing of sputum, and nutritional support to promote healing, improve electrolyte imbalance, and prevent malnutrition due to chronic bile loss. Surgical management includes repair of the diaphragmatic rent, repair of the fistulous tract, and dissection of the damaged lung tissue with lobectomy in most cases. It can be done either through video-assisted thoracoscopic surgery(VATS) as done in our case or through thoracotomy [1,4,7,8]. Regular follow-up to monitor clinical status, lung functions, and nutritional status to ensure post-operative healing is necessary.

CONCLUSION

This case of Broncho-biliary fistula in a 4-year-old child underscores the complexity and diagnostic challenges of this rare condition in pediatric patients. A gross examination of sputum for the sought-after diagnosis should be done. Early identification and prompt treatment results in positive outcomes and enhanced quality of life in patients.

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