

mitosis, necrosis, or atypical stromal component was not present. The final diagnosis was a parosteal lipoma of the rib.

After the surgical resection of the mass, the patient showed no postoperative complications, including local recurrence during the follow-up period of 4 months.

Comment

Parosteal lipomas are relatively rare and usually asymptomatic benign lesions composed mainly of mature adipose tissue [4]. The incidence of parosteal lipoma is 0.3% of all lipomas [4, 5]. Although the possibility of mixed origin due to foci of metaplastic cartilage undergoing chondral ossification within adipose tissue and traumatic origin has been suggested [6, 7], the cause of parosteal lipoma is still unknown. In our case, the patient had a previous history of trauma in the tumor site.

Age distribution of patients with common parosteal lipoma ranges from 40 to 60 years old. Patients with parosteal lipoma of the rib range in age from 57 to 60 years old [4–6]. Most patients complain a history of a slow-growing, large, painless, and nontender mass. Sometimes motor and sensory disturbances from adjacent nerve compression may occur [8, 9]. In our case, the patient was 50 years old and did not complain of the neurologic symptom.

The radiologic features of parosteal lipoma are characterized by osseous reactive changes, such as bowing of bone or erosion of bony cortex secondary to the adjacent lipomatous tumor. The findings of a computed tomographic scan for parosteal lipoma include a variable degree of surface bone productive changes, ranging from subtle to conspicuous thickening of cortex and various sized ossifying processes or excrescent bone spicules [4, 5].

On gross and microscopic examinations, the parosteal lipomas have strongly adhered to the underlying periosteum of bone and well-demarcated greasy, yellowish, soft masses encapsulated by thin fibrous membrane. These tumors consist of mature adipose tissue identical to the other lipomas of soft tissue. Foci of osseous metaplasia, cartilage, and osseous excrescences or thickening of cortex extending from and attaching the lesion to the surface of bone are commonly observed [10].

The primary treatment of parosteal lipoma is complete surgical excision. If nerve entrapment accompanies, the tumor will be eliminated as soon as possible before irreversible atrophic change of muscle. The parosteal lipomas strongly adhere to the underlying periosteum of affected bone. Therefore, partial resection of involved bone may be required. Local recurrence is uncommon and malignant transformation has not been documented yet [5].

In brief, we have described an extremely rare case of parosteal lipoma of the rib. Although the incidence is extremely rare, parosteal lipoma should be considered in the radiologic and pathologic differential diagnosis.

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Surgical Experience in a Baby With Congenital Broncho-Biliary Fistula

Ayla Günlemez, Melih Tugay, Levent Elemen, MD, Gülcan Türker, Nagihan İnan Gürcan, Hakan Demir, Yeşim Gürbüz, and Tulay Hosten

Departments of Pediatrics, Neonatal Intensive Care Unit, Pediatric Surgery, Radiology, Nuclear Medicine, Pathology, and Anesthesiology, Kocaeli University School of Medicine, Kocaeli, Turkey

Congenital tracheobiliary and bronchobiliary fistulae are rare malformations in which patent communications exist between the respiratory system and biliary tract, respectively. We present a newborn who was admitted with respiratory distress and bilious tracheal discharge. Investigation revealed a bronchobiliary fistula originating from the left main bronchus, as well as biliary atresia. Excision of the bronchobiliary fistula was successful and the connection between biliary tract and gastrointestinal system was established by performance of a Roux-en-Y cholecysto-jejunostomy. Diagnostic tools, differential diagnosis, and surgical correction strategies are discussed.

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Congenital respiratory system biliary tract fistula is a rare developmental anomaly. It has been reported in conjunction with other congenital anomalies, such as

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Address correspondence to Dr Tugay, Kocaeli Universitesi Tıp Fakultesi Hastanesi, Çocuk Cerrahisi AD. Kat: 2, Umuttepe, Kocaeli, 41380, Turkey; e-mail: tugaym@hotmail.com.



Fig 1. Preoperative multi-detector computed tomographic scan shows the left broncho-biliary fistulae (arrow) coursing from the left main bronchus to the left hepatic lobe.

biliary tract malformations, diaphragmatic hernia, and esophageal atresia [1–3]. Various types of tracheobiliary or bronchobiliary fistulae (BBF) have been described [4].

A 9-day-old female infant was admitted with respiratory distress and bilious vomiting. Her medical history disclosed that the patient had experienced bilious vomiting and subsequent respiratory distress on the postnatal third hour. Physical examination revealed yellow colored salivation and diminished lung sounds on the left hemithorax. Chest roentgenogram showed total left lung consolidation. On day 2 of her admission, the baby was endotracheally intubated due to left lung atelectasis complicating with respiratory distress. As bilious fluid discharge started to come out continuously from the endotracheal tube, the patient was submitted to multi-detector computed tomographic scan (MDCT) and Tc-99m hepatobiliary iminodiacetic acid (HIDA) scintigraphy. After the MDCT axial and coronal images were reformatted (Fig 1), a virtual bronchoscopy was performed that demonstrated a fistula coursing from the left main bronchus to the left hepatic lobe (Fig 2). Hepatobiliary scintigraphy revealed radio-tracer runoff in the left lung and dissemination through the right lung without radionuclide passage to gastrointestinal system. Thus, the patient was taken to operation with the presumptive diagnosis of a left BBF. In the operation, the BBF tract opening was located at the entrance of the left main bronchus by bronchoscopy. Then, a right posterolateral thoracotomy was performed from the fourth intercostal space. The bronchobiliary fistula was divided and ligated at the level of the tracheal entrance with nonabsorbable sutures (5-0 Prolene [Ethicon, Edinburgh, United Kingdom]). Explorative laparotomy revealed absence of common bile duct with a normal looking gallbladder. Intraoperative cholangiography was performed, and biliary tract connection to the gastrointestinal system was not demonstrated.

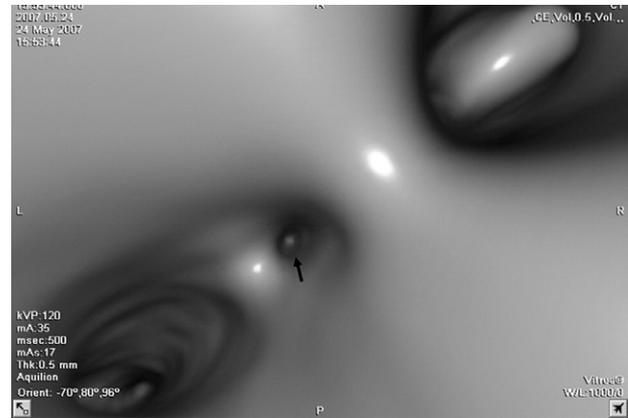


Fig 2. Virtual bronchoscopy image shows origin of broncho-biliary fistulae (arrow) in left main bronchus.

Instead, contrast material filled up the intrahepatic biliary system, thus confirming the absence of the common bile duct. Roux-en-Y cholecysto-jejunostomy was used to resolve the biliary obstruction. Although biliary drainage ceased, and left lung atelectasis recovered on the postoperative early period, bile stained sputum recurred on postoperative day 3. The patient once more went through MDCT, and a thoracic cyst over the diaphragm was demonstrated (Fig 3). Hepatobiliary scintigraphy showed radio-tracer collection in the left lung and normal biliary drainage to the gastrointestinal system. She underwent a re-thoracotomy through the sixth intercostal space that demonstrated previously divided fistula ends were apart without connection in between. Further exploration of the posterior mediastinum revealed a cyst sited next to the esophagus, communicating with the bronchial tree and extending from the esophageal hiatus through the tracheal bifurcation with dimensions of 25 × 40 mm. After the aspiration of bile and



Fig 3. Postoperative multi-detector computed tomographic scan shows the remnant sac (arrow).

further dissection, the cyst was excised with the distal ligation site at the diaphragm.

Recovery was uneventful and the patient was discharged on postoperative day 5. She is still doing well without any complications for a follow-up period of 9 months.

Histopathologic examination of the resected specimens in both of the operations revealed that they were lined by ciliated pseudostratified epithelium surrounded by a smooth muscle layer. Also there was a cartilage islet surrounded by mucinous glands.

Comment

Congenital tracheobiliary or BBF is an uncommon anomaly. Although the majority of these cases have been seen in neonates, age distribution may vary from infants to adults [1–7]. Various localizations of BBF have been described with only two on the left side [4]. Biliary vomiting, which is frequently misdiagnosed as gastroesophageal reflux, and respiratory distress are the most frequent symptoms in newborns [1, 2, 4]. In the presented case, a high amount of clear bilious discharge from the endotracheal tube was the key symptom. Bilious vomiting in the absence of intestinal obstruction should be indicative for the anomaly [4].

Two embryological explanations have been advanced for the origin of the BBF. Neuhauser and colleagues [4], the first authors to describe this malformation in 1952, believed that the anomaly resulted from a duplication of the upper gastrointestinal tract. In 1957, Bremer [4], and then in 1966, Sane and colleagues [1, 4], suggested that presence of a communication between an anomalous bronchial bud and an anomalous bile duct would be explanatory. It is evident, as in our case, that the histologic features of the reported fistulas in the literature support the second theory [4].

Diagnostic studies are important for the surgery plan. In the present case, MDCT and virtual bronchoscopy demonstrated the anatomy of the anomaly. Hepatobiliary scintigraphy revealed biliary drainage to the tracheobronchial tree, as well as lack of connection between the biliary tract and the gastrointestinal system, which was also a clue for the anomaly. Because common bile duct atresia is the most common associated anomaly [1, 4, 6], we believe that performance of hepatobiliary scintigraphy is crucial for defining the accompanying biliary tract malformations [2, 4]. Although intraoperative bronchoscopy demonstrated the fistula opening, we believe bile staining of the tracheobronchial tree could make this difficult. For this particular reason, performance of virtual bronchoscopy seems to be beneficial in showing the anatomy in better details compared with conventional bronchoscopy.

We performed thoracotomy from the fourth intercostal space in the first operation to stay close to bronchial opening. Despite the ligation of the BBF, symptoms recurred and further radiological evaluation and reoperation were required. We assume that this condition was due to transitional obliteration of bile outlet leading to pressure increase in biliary tract causing postoperative

opening of a fistula beyond the ligation place, which was not apparent in the first operation. This fistula could not be demonstrated in the intraoperative cholangiography, as it was not patent. Cessation of bilious discharge after the first operation might be suggestive for our hypothesis. On the other hand, it is possible that the cyst was present and missed in the first MDCT. As the computed tomographic images were taken in a little oblique manner, the cyst might partially overlay the density of the left lung leading to insufficient interpretation. Another possibility is that the lesion was missed at the first operation due to inadequate exposure. Therefore, we strongly recommend thoracotomy from the sixth intercostal space, which provides better exposure. Dissection of the BBF all the way down through the diaphragmatic hiatus and total excision is required rather than division and ligation of the fistula.

As the fistula is the only bile outlet after BBF excision, biliary tract connection to the gastrointestinal system should be established. Roux-en-Y choledochojejunostomy can be performed for this purpose [4]. A fistulo-enteric anastomosis has also been described, and potentially could have been done at the region of the diaphragm [4]. Porto-enterostomy may be another choice in case of an accompanying extrahepatic biliary atresia. If normal gallbladder exists, drainage of bile through cholecysto-enteric anastomosis can also be performed [1]. In the present case, we think that performance of Roux-en-Y cholecysto-jejunosotomy was technically easy and appropriate by shortening operation time. The intra-abdominal portion of the fistula was remained untouched, not to cause problems, as it is recommended [1].

In conclusion, clear yellow-stained sputum or vomiting should alert the physician for the possibility of connection of the respiratory system and biliary tract. The MDCT and virtual bronchoscopy are the most important diagnostic tools for the detailed description of the anomaly. Hepatobiliary scintigraphy is crucial for defining the accompanying biliary tract malformations. An operation should be performed through the sixth intercostal space, and total removal of the BBF is mandatory.

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