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Case Report

Bronchogenic Cyst: A Rare Cause of a Retroperitoneal Mass in a Neonate

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ABSTRACT

***Background*:** Bronchogenic cysts (BCs) are congenital abnormalities of the primitive foregut, which are commonly found in the lung and mediastinum; however, they may rarely occur in the retroperitoneal region.

Case report: We present a case of 20-day-old male newborn with a prenatal diagnosis of an abdominal mass who was referred and admitted to our hospital for further evaluation. Abdominal ultrasound and computed tomography scan demonstrated a retroperitoneal ill-defined solid mass locating on the upper ridge of the left kidney in close contact with the posterior wall of the stomach and pressure effects on the spleen hilum. Concerning presumptive diagnosis of neuroblastoma, less likely Wilms tumor, and subdiaphragmatic sequestration, surgery was carried out, and the mass removed completely. Pathological examination revealed a multiloculated cystic-solid mass with cystic spaces which was lined by respiratory epithelium, and the underlying fibrotic stroma contained islands of mature cartilage consistent with the diagnosis of bronchogenic cyst.

Conclusion: Although BCs rarely occur, they should be considered in the differential diagnosis of a retroperitoneal mass in the neonates.

Keywords: Bronchogenic cyst, Congenital abnormalities, Neonate, Retroperitoneal mass

Introduction

Bronchogenic cysts (BCs) are benign congenital malformations of the tracheobronchial tree during embryonic development. They are usually located in the thorax within the mediastinum or in the lung parenchyma; however, they can rarely be observed in extrathoracic locations (1). It should be noted that the retroperitoneal location is extremely rare (2, 3).

The BC was first reported by Meyer et al. (1859), and the possible occurrence of retroperitoneal BCs was first mentioned by Miller et al. in 1953 (2). About 66 cases of retroperitoneal BCs have been reported in the English literature so far (3).

The BCs are usually asymptomatic, and the clinical symptoms occur as a result of secondary infection, bleeding, perforation, or compression of adjacent vital structures (4, 2). The location and type of BCs determine the form and severity of the symptoms (5). Retroperitoneal BCs are usually diagnosed incidentally (2).

To the best of our knowledge, this is the first report of a congenital retroperitoneal BC diagnosed and treated successfully in the neonatal period in Iran.

Case report

A 20-day-old male neonate with a gestational age of 40 weeks born by cesarean section from nonconsanguineous parents was referred to our hospital because of an abdominal mass in the left adrenal region which had been discovered in the 37th week of his gestational age by ultrasound. Pregnancy and birth history were uneventful.

The infant was admitted for further evaluation. The first physical examination was unremarkable.

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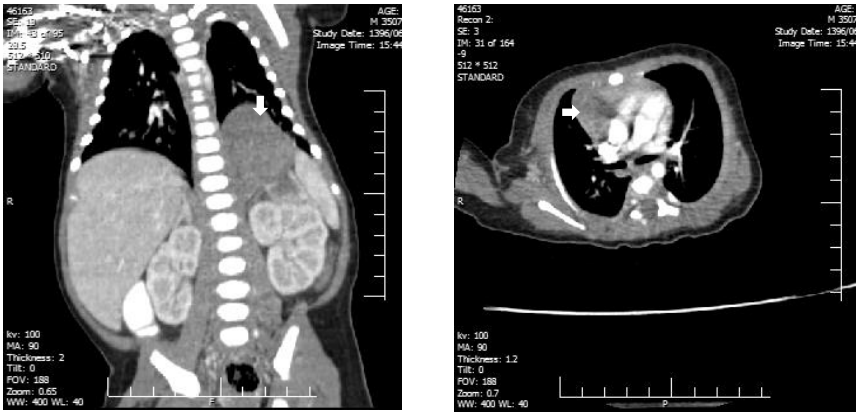

Figure 1. Axial and spiral computed tomography scan with contrast media

No organomegaly or palpable mass was detected on abdominal examination. Lab data, including complete blood count, biochemistry, and liver function tests were within normal limit. During hospitalization, he had few episodes of fever. Sepsis workup was performed, and the administration of antibiotics improved his general condition. It is worth mentioning that the cultures were negative.

No remarkable findings were noted on the chest radiograph. Abdominal sonography revealed a 46×34 mm retroperitoneal ill-defined solid mass on the upper ridge of the left kidney close to the hilum of the spleen. The mass had vascular flow and did not pass the midline. Based on its homogeneity and echogenicity, it resembled neuroblastoma and less likely Wilms tumor.

Vanillylmandelic and homovanillic acids were 0.4 (Normal range:0.169-1.351mg/24 h) and 0.43 mg (Normal range: 1.4-4.3mg/24h), respectively, in 24 h collected urine. The serum level of alfa fetoprotein and beta human chorionic gonadotropin was normal as well.

Spiral abdominopelvic computed tomography (CT) scan with contrast media demonstrated a 50×37×32 mm soft tissue mass in the left suprarenal region. The mass was in close contact with the posterior wall of the stomach and spleen. It had displaced the adrenal gland laterally and left diaphragm superiorly. Moreover, there was a thin branch from aorta supplying the mass. All the other organs were normal (Figure 1). Neuroblastoma and less likely subdiaphragmatic sequestration were suggested in the differential diagnosis of the lesion by the radiologists.

Appropriate consultations, including hematology, surgery, and cardiology were performed for our patient, and he underwent surgery for either biopsy or removal of the whole mass.

The surgery was performed under general anesthesia. The mass had a retroperitoneal origin and was located on the top of the adrenal gland with a distinct and irregular border measuring 5.5×4×2 centimeter. The mass was removed completely, the diaphragm reconstructed, and a tube was inserted for drainage.

The histopathologic examination showed a solid cystic mass with a soft whitish surface and smooth consistency. In the microscopic examination, a hamartomatous tissue was noted which was mostly composed of small and large multiloculated cystic spaces lined by respiratory epithelium. The intervening stroma was cellular and composed of fibromuscular tissue with foci of mature cartilage indicating a bronchogenic multiloculated cyst. No other types of tissues were observed ruling out the diagnosis of teratoma (Figure 2).

The patient had an uneventful recovery and discharged with a very good general condition. The patient was clinically well and free of complications during a-9-month follow-up,

Discussion

The BCs with an incidence of 1 per 100,000 population (6) are rare congenital malformations originated from anomalous budding of the embryonic foregut, which usually detected above the diaphragm, especially in the mediastinum or in the pulmonary parenchyma (2, 3, 7). A few cases of bronchogenic cyst below the diaphragm are reported in the literature. Their occurrence is very rare in the retroperitoneum, and the mechanism is not clear (2). It has been postulated that before the fusion of diaphragm, the aberrant bud detaches from the primitive foregut and migrates toward the abdomen resulting in a retroperitoneal bronchogenic cyst. Another hypothesis is the possibility of intraabdominal

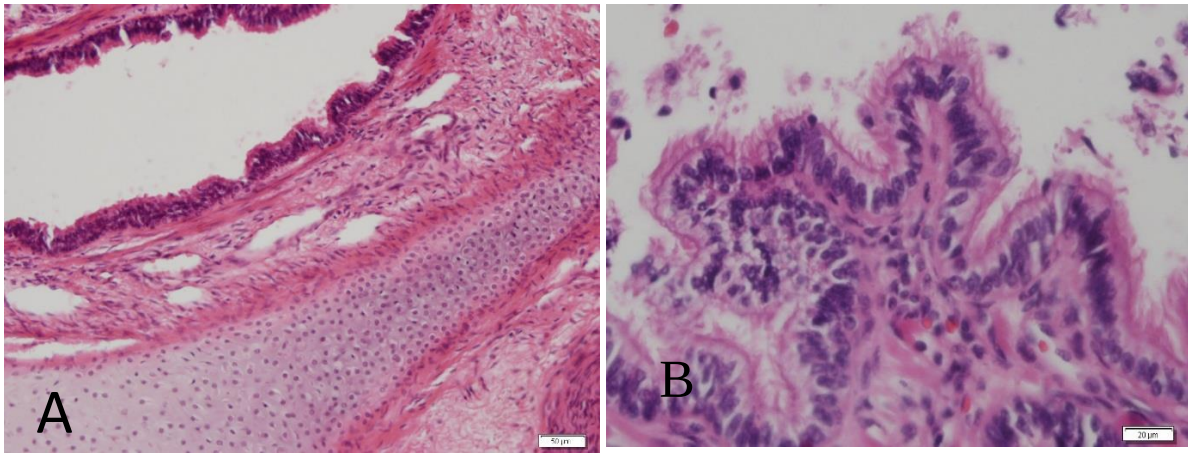

Figure 2. Pathology slides: Histopathology examination showing cystic spaces that are lined by respiratory type epithelium. The underlying stroma was composed of fibromuscular tissue with foci of mature cartilage (hematoxylin and eosin stain; A: X250; B: X800)

budding from the foregut which seems less reliable (2, 3).

Kizu et al. described 39 BCs with the intraabdominal origin of which 25 cases were in retroperitoneal origin (8). Among eight children with intraabdominal lesions, six cases were in the retroperitoneal area (9). The majority of these cysts are located in an imaginary triangle envisioned posterior to the stomach, defined by the midline, splenic vein inferiorly, and the diaphragm superiorly (2, 10). According to the literature, more than 80% of the retroperitoneal BCs are located near the left adrenal gland (2, 3). In our patient, the cyst was located at the top of the left adrenal in close contact with the posterior wall of the stomach and spleen. The second most common location is the peripancreatic region (2).

The finding of a retroperitoneal mass in neonates usually raises various differential diagnoses, such as neuroblastoma, adrenal hemorrhage, Wilms tumor, teratoma, and subdiaphragmatic sequestration (11). Imaging modalities, such as X-ray, ultrasound, CT scan, and magnetic resonance imaging are the methods for diagnosis; however, the exact preoperative diagnosis is difficult, and only histologic examination can confirm the definite diagnosis.

Our patient was a male infant, retroperitoneal BCs occur with equal frequency in both males and females with a wide age range (3). A male predominance of 4:1 was stated by Maly et al. (10). According to a study conducted by Ubukata et al., an age range of 3 weeks to 81 years have been reported in this regard (12). In most instances, BCs are asymptomatic and are detected incidentally (2, 10). Prenatal diagnosis of BCs might be possible and could prevent potential complications (13). Although the prenatal diagnosis of thoracic BCs is increasing, this diagnosis is difficult for abdominal lesions. Bagolan et al. reported the prenatal ultrasound observation of an intra-abdominal bronchogenic cyst for the first time (9).

The treatment of retroperitoneal BCs is surgical removal. The excision is recommended to establish a definite diagnosis, alleviate any symptoms, and prevent complications (2, 12). Some reports of laparoscopic excision of retroperitoneal BCs have been reported in the literature (9, 14); however, there is limited experience among neonates in this regard.

When the diagnosis of BCs is suspected, minimally invasive procedures, such as endoscopic ultrasonography-guided fine needle biopsy aspiration and endoscopic submucosal dissection should be considered. They are valuable diagnostic methods in both adults and children; however, it is more difficult in small children (9). The prognosis of the bronchogenic cyst is favorable; nonetheless, the appropriate follow-up is needed for patients.

Conclusion

Despite the rarity of retroperitoneal BCs, it should be considered in the differential diagnosis of retroperitoneal masses among neonates, especially when they are found in the left upper quadrant.

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Conflicts of interests

The authors declare that they have no conflicts

of interest.

Ethical Considerations

Written informed consent was obtained from parents to report this case.

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