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Reporte de Caso

Perinatal results of conservative management in fetal bronchopulmonary sequestration. Report of a case

Resultados perinatales del manejo prenatal conservador en secuestro broncopulmonar fetal. Reporte de un caso

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RESUMEN

Introduction: Congenital bronchopulmonary malformations are rare but potentially fatal anomalies in fetuses and newborns. They require urgent diagnostic evaluation from the prenatal and neonatal period. In 5.9% of cases, the lesion resolves spontaneously. The scarcity of evidence on its natural evolution has generated divergences in the management strategies of prenatal cases and of asymptomatic neonates, which results in an always controversial issue. Prenatal regression is common in cases diagnosed before birth, and most children without symptoms beyond the neonatal period will remain symptom-free throughout infancy. The lack of statistical information in our country, and in our center, and the scarcity of comparative studies on the different therapeutic methods available, make it difficult to formulate unique prenatal therapeutic protocols. We present a clinical case attended from the prenatal period at the University Hospital of Guayaquil in the year 2023 with expectant management of the fetus and resolution of maternal comorbidities with favorable perinatal outcome. Of note is the spontaneous regression from mild polyhydramnios to severe oligohydramnios in 4 weeks,

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simultaneous with the resolution of the fetal pleural effusion and the rearrangement of the structures of the fetal mediastinum, previously displaced by the tumor and the tension hydrothorax. **Clinical case:** A 32-year-old multiparous patient detected late with a pregnancy of 30.1 weeks of gestation complicated by maternal syphilis and fetal bronchopulmonary sequestration (BPS) with pleural effusion and severe displacement of the fetal mediastinum to the right, without fetal hemodynamic compromise. Diagnoses confirmed by obstetric ultrasounds, serial fetal Doppler and fetal MRI. It evolves with mild polyhydramnios and is managed expectantly until 37 weeks when, after spontaneous resolution of the hydrothorax, it progresses to severe oligohydramnios and is completed by segmental cesarean section. The neonate receives a thoracotomy with resection of the lung mass at 7 days after birth and the diagnosis is confirmed by pathology. **Conclusions:** We present a clinical case of a rare fetal pathology in all its types worldwide and underdiagnosed in our environment, treated late at 30.1 weeks of gestation with multiple maternal-fetal comorbidities with spontaneous improvement in the prenatal period, largely managed expectantly. due to the diagnostic and therapeutic challenges faced, in the absence of a fetal medicine and surgery service in the area. Despite this, expectant fetal management is an internationally valid option, with very favorable results in most cases without hydrops or other signs of fetal heart failure. Comparative studies of different prenatal management methods are lacking partly due to the rarity of the pathology; In our environment, its dispersion and late prenatal diagnosis with poor accessibility to subspecialists in maternal-fetal medicine and fetal surgery can negatively influence perinatal outcomes..

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Keywords: Bronchopulmonary sequestration (BPS); Congenital adenomatous cystic airway malformation (CAM); Congenital pulmonary malformation (CPM); preserved treatment; hybrid lesions; prenatal.

ABSTRACT

Introducción: Las malformaciones broncopulmonares congénitas son anomalías raras pero potencialmente fatales en fetos y recién nacidos. Requieren evaluación diagnóstica urgente desde el período prenatal y neonatal. En el 5,9% de los casos, la lesión se resuelve espontáneamente. La escasez de evidencia sobre su evolución natural ha generado divergencias en las estrategias de manejo de los casos prenatales y de los neonatos asintomáticos, lo que resulta en un tema siempre controversial. La regresión prenatal es común en los casos diagnosticados antes del nacimiento, y la mayoría de los niños sin síntomas más allá del período neonatal permanecerán asintomáticos durante toda la infancia. La falta de información estadística en nuestro país, y en nuestro centro, y la escasez de estudios comparativos sobre los diferentes métodos terapéuticos disponibles, dificultan la formulación de protocolos terapéuticos prenatales únicos. Presentamos un caso clínico atendido desde el período prenatal en el Hospital Universitario de Guayaquil en el año 2023 con manejo expectante del feto y resolución de las comorbilidades maternas con resultado perinatal favorable. Destaca la regresión espontánea de polihidramnios leve a oligohidramnios severo en 4 semanas, simultáneo con la resolución del derrame pleural fetal y el reordenamiento de las estructuras del mediastino fetal, previamente desplazadas por el tumor y el hidrotórax a tensión. **Caso clínico:** Paciente multípara de 32 años detectada tardíamente con embarazo de 30.1 semanas de gestación complicado con sífilis materna y secuestro broncopulmonar (SBP) fetal con derrame pleural y desplazamiento severo del mediastino fetal a la derecha, sin compromiso hemodinámico fetal. Diagnósticos confirmados por ecografías obstétricas, Doppler fetal seriado y resonancia magnética fetal. Evoluciona con polihidramnios leve y se maneja expectante hasta las 37 semanas cuando, tras resolución espontánea del hidrotórax, progresa a oligohidramnios severo y se completa mediante cesárea segmentaria. El neonato recibe una toracotomía con resección de la masa pulmonar a los 7 días de nacido y el diagnóstico se confirma por patología. **Conclusiones:** Presentamos un caso clínico de una patología fetal rara en todos sus tipos a nivel mundial y subdiagnosticada en nuestro medio, tratada tardíamente a las 30.1 semanas de gestación con múltiples comorbilidades materno-fetales con mejoría espontánea en el periodo prenatal, manejada en gran medida de manera expectante debido a los retos diagnósticos y terapéuticos que se enfrentan, en ausencia de un servicio de medicina y cirugía fetal en la zona. A pesar de ello, el manejo fetal expectante es una opción válida a nivel internacional, con resultados muy favorables en la mayoría de los casos sin hidrops ni otros signos de insuficiencia cardiaca fetal. Faltan estudios comparativos de diferentes métodos de manejo prenatal en parte por la rareza de la patología; en nuestro medio, su dispersión y diagnóstico prenatal tardío con poca accesibilidad a subespecialistas en medicina materno-fetal y cirugía fetal pueden influir negativamente en los resultados perinatales..

Palabras Clave: Secuestro Broncopulmonar (BPS), Malformación quística adenomatosa congénita de las vías respiratorias (CPAM), Malformación Pulmonar Congénita (CPM), tratamiento preservado, lesiones híbridas, prenatal.

Introduction

Congenital bronchopulmonary malformations are rare, but potentially fatal anomalies in fetuses and newborns, and require urgent diagnostic evaluation from the prenatal and neonatal period, which is easily performed by obstetric ultrasound in the first trimester as the method of choice[1,2]. Bronchopulmonary sequestration (PS), congenital cystic adenomatous malformation (CCAM), congenital lobar emphysema (CLE) and bronchogenic cyst (BC) are the four main congenital cystic lesions, but they share similar embryological and clinical characteristics[3,4]. Defined as a heterogeneous group of solid intrathoracic pulmonary masses that cause lesions of dysfunctional lung tissue and that in the case of bronchopulmonary sequestration will be supplied by an arterial vessel branch of the thoracoabdominal aorta 5, generally easy to visualize by color Doppler and being more difficult to detect . evaluate its venous drainage even by fetal MRI. The Great Ormond Street Hospital in London described one of the largest series to date on these pathologies in 2017. They analyzed the long-term evolution of 119 cases treated at the institution (18 years). Of these, 43% (51 patients) underwent surgery, mainly due to respiratory infections. 57% (68 patients) received conservative treatment for an average of 9.9 years. In 5.9% (4 patients), the lesion resolved spontaneously. 76% (52 patients) continue to be followed up and remain asymptomatic, without presenting malignancy[6].

The scarcity of evidence on the natural evolution of these congenital respiratory tract malformations has resulted in a divergence in the management strategy of asymptomatic cases, making it undoubtedly a controversial topic[6,7]. Prenatal regression is common in cases diagnosed prenatally, 5 and the majority of children who do not present symptoms beyond the neonatal period will remain symptom-free throughout their childhood, with hemoptysis and respiratory infections being the main causes of active behaviors. Future analysis with longer follow-up could provide new insights to identify children at risk of developing symptoms.

In our country, statistical information in this regard is insufficient and is often dispersed in small series, mostly postnatal studies of pediatric surgery. In 2022, a group of pediatric surgery residents at the Catholic University of Santiago de Guayaquil mention that they did not find previous studies that relate 2 therapeutic methods, such as lesion resection vs. endovascular or percutaneous embolization 8 . Until 2019, Cruz et al 5 carried out a review in which of 150 articles from various databases they chose only 29 in which a combination of prenatal diagnosis and fetal treatment was found, which justifies the difficulty in stating prenatal therapeutic protocols. unique.

Clinical case

Information of the Patiente

We present a 32-year-old, multiparous patient with 4 births and 3 previous abortions and intergenic period of 4 years, with a history of syphilis since her second pregnancy, having received incomplete treatment on 2 occasions. Received due to a clinical picture of threatened preterm labor secondary to vaginosis and recurrent urinary tract infection during pregnancy; At 30 weeks with 1/7 day of gestation, with insufficient prenatal controls and obstetric ultrasounds, a diagnosis of fetal left pleural effusion was recorded.

Clinical Findings

Afebrile, hydrated, hemodynamically stable patient given BP: 101/73 mmHg, HR: 90 bpm, RR: 21 rpm, O2 saturation: 99%, Temperature: 36.3°C and alert level of consciousness. Size: 153 cm, weight: 50 kg. The obstetric physical examination revealed uterine height: 33 cm, single fetus, longitudinal left dorsum, cephalic, FHR: 156 bpm, regular uterine dynamics: 3/10 minutes/45 seconds/ baseline 10 mmHg/85 mmHg. Erythematous external genitalia with yellow-green leucorrhoea, thick, fluid, fetid, abundant, also con-

firmed by speculoscopy where amniorrhea did not appear with any maneuver through the OCE of the cervix of the multiparous woman presented to us.

Diagnostic Evaluation

Clinical laboratories:

Leukocytes 14300, neutrophils 78%. Reactive syphilis, VDRL positive with 1/16 dilutions, non-reactive HIV. Pathological urinalysis with abundant leukocytes (uncountable) and positive nitrites with pH>7.3. Glucose: 73 mg/d . Direct examination of vaginal discharge not available. Negative emergency crystallography.

Emergency obstetric ultrasound:

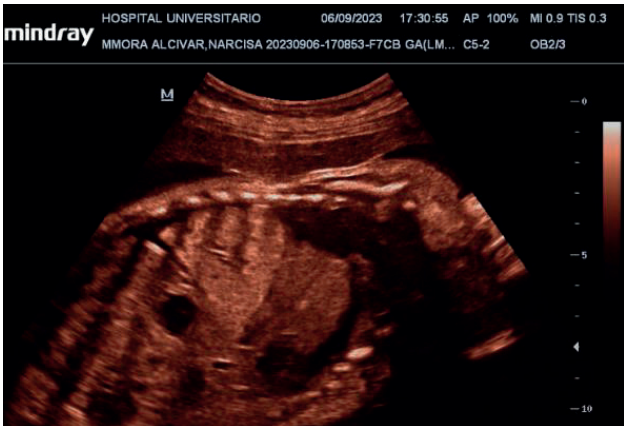


Image 1. Solid mass in left fetal hemihtorax in a sagital plane. B mode.

Single, longitudinal, live fetus, left anterior dorsum, high and mobile head, active and reactive fetal movements. FHR: 153 bpm. Male sex. DBP: 62.4mm. DC: 294.9 mm. DOF: 100.6 mm. AC: 298.3mm. LF: 62mm. DTC: 42.5mm. CM: 8.3 mm.DC/AC: 0.99,

LF/AC: 20.78. PFE: 1957 grams. EG/US: 32 weeks and 6/7 days. ILA 21.8 cms that progresses to 24 cms at 2 weeks, and MBV: 9.1 cms. Ultrasound finding: a slightly hyperechogenic mass is seen that occupies the basal 1/3 of the left hemithorax, with regular borders of 43.9 mm by 42.0 mm (Image 1 and 2), without macroscopic cystic images in its echo structure, which is irrigated by a large-caliber nutrient vessel branch of the descending thoracic aorta (Illustration 3).

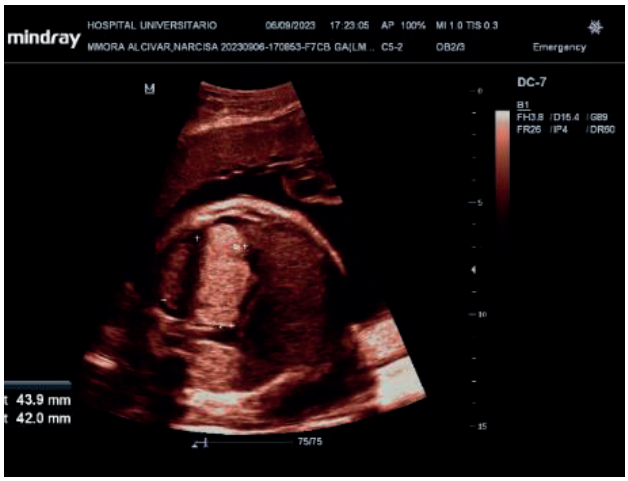


Image 2. Solid mass in fetal left hemithorax. Corte transversal. B mode.

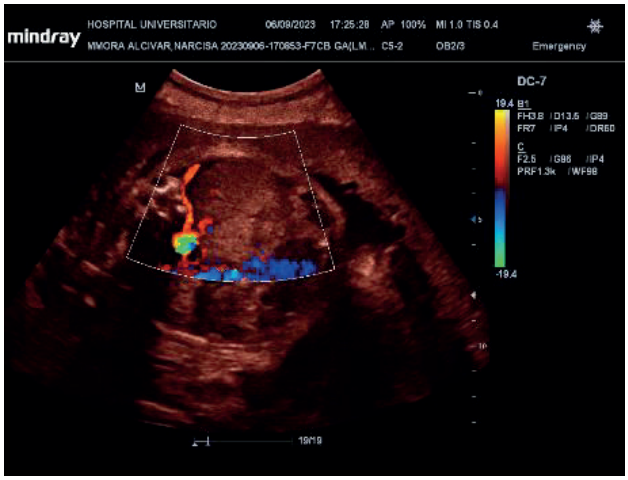
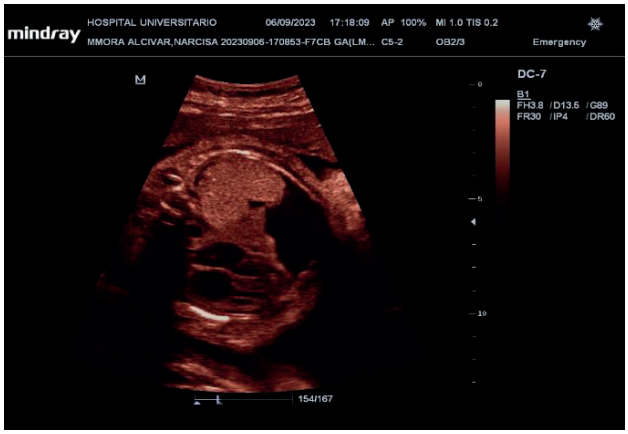


Image 3. Central artery. Transvers plane. Doppler color.



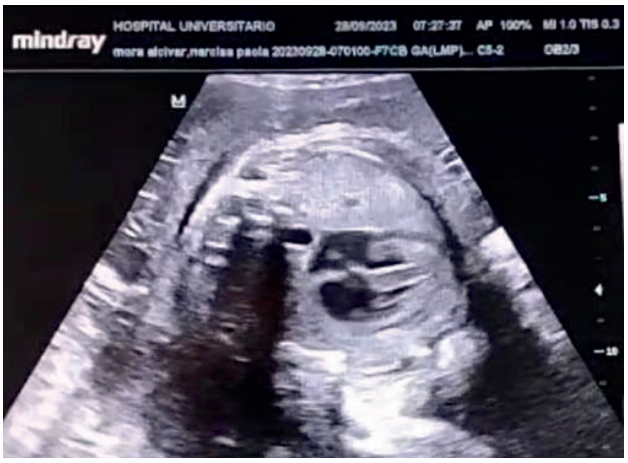


Image 4. Fetal bronchopulmonary sequestration with pleural effusion.

Therapeutic Intervention

We began prescribing empirical vaginosis therapy with clindamycin 100 mg vaginally (suppositories) QD, for 6 days followed by lactobacilli acidophilus (Vaginal Tablets 0.03 Mg) for 6 days. Additionally, Fosfomycin 3 grams orally in a single dose is indicated while awaiting urine culture results. Similarly, nifedipine 10 mg PO TID for 72 hours and progesterone 200 mg PO, QD, for 10 days are prescribed. We evaluated the patient weekly with partial improvement and even without urine culture results, it was decided to administer cephalexin 500 mg PO, QD for 7 days. During this time, the patient's referral to the National Center for Maternal Fetal Medicine in the city of Quito is coordinated. The patient does not receive the complete medication prescription despite having it available, nor does she go to the referral center, presenting irregular evolution with recurrence of uterine dynamics. In addition, a complete induction scheme of fetal lung maturation is placed with betamethasone 12 mg IM, QD for 2 doses.

At 33.1 weeks of gestation, she was admitted to the emergency room with regular intense uterine contractions that initially did not improve with rehydration or the administration of tocolytics at the emergency therapeutic doses indicated, but then while waiting for a surgical appointment, the dynamics subsided with nifedipine PO 3 doses. (10mg/10mg/20mg) and magnesium sulfate IV for 12 hours, which led to his transfer to the High Obstetric Risk room, where he received intravenous treatment with ampicillin 1g QID for 7 days and clindamycin 100 mg vaginally for 6 days, improving substantially. . Due to the obstetric, psychosocial and perinatal risk, they remain hospitalized awaiting referral to the National Center for Fetal Medicine in the city of Quito. During this time, the therapeutic regimen for syphilis was completed with benzathine penicillin 2.4 million IM weekly for 3 weeks with subsequent serological control reporting reactive syphilis, but with non-reactive VDRL and dilutions of 1/6 (initially 1/16).

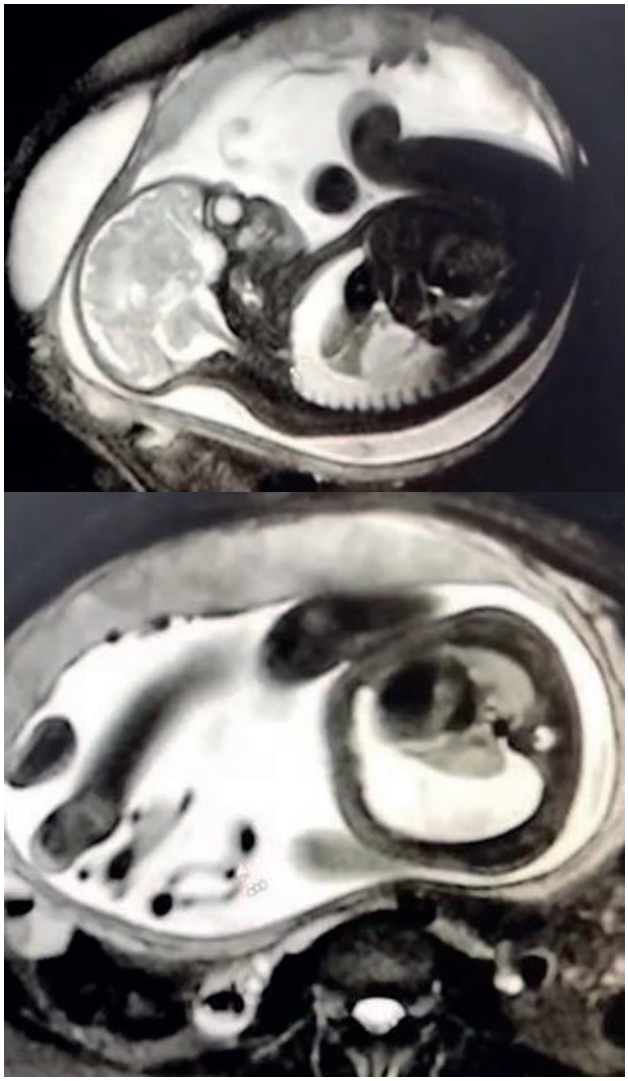


Image 5. Fetal MRI: Pleural effusion in the case of fetal bronchopulmonary sequestration (left: Transverse section), partial spontaneous reabsorption (sagittal section).

Termination of Pregnancy and Neonatal Evolution

At 37 weeks of gestation due to severe oligohydramnios (ILA: 2 cm / MBV: 1 cm), it was decided to terminate the pregnancy electively via segmental cesarean section

in which the absence of amniotic fluid was confirmed, obtaining a live male newborn with Apgar 8- 9 at one minute and 5 minutes, does not require stimulation and transitions to attachment and breastfeeding in the immediate postpartum period without complications. During hospitalization, a simple chest tomography was performed on the neonate, revealing a solid mass in the posterior segments of the left lower lobe (Illustration 5).

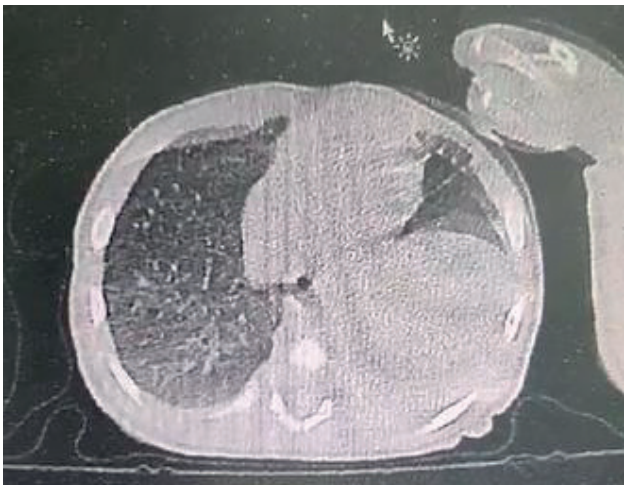


Image 6. Neonatal CT showing an intrathoracic mas with suspected bronchopulmonary sequestration (Transverse section).

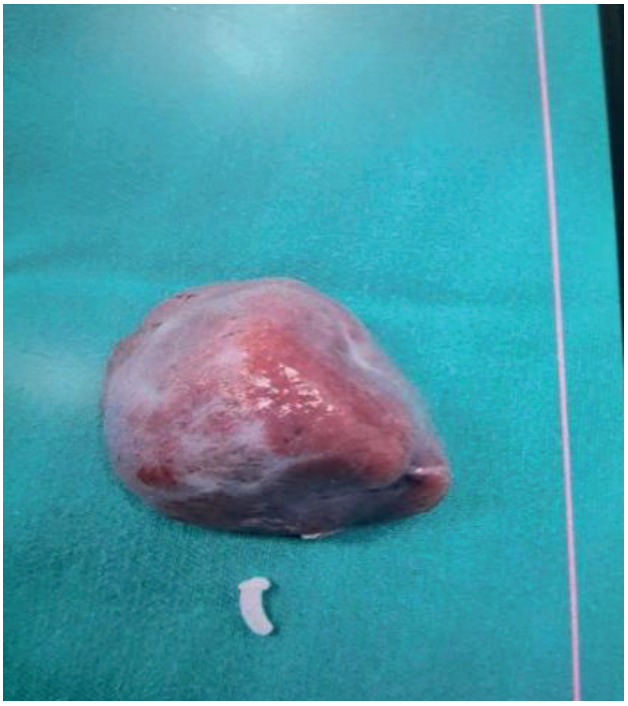


Image 7. Anatomical piece, extralobar bronchopulmonary sequestration.

Due to bronchopulmonary infection and acute respiratory distress, the neonate was transferred to neonatal intensive care at the Dr. Roberto Gilbert Elizalde Children's Hospital of the Guayaquil Charity Board. After stabilizing the patient ventilatorily and hemodynamically, it was decided to perform a thoracotomy and resection of the mass, after which a successful recovery was obtained and the patient was discharged from the institution 7 days after the surgical procedure without presenting other complications (Illustration 6 and 7). And finally, the result of the histopathological study is obtained that confirms the initial suspicion of extralobar bronchopulmonary sequestration of the lower lobe of the left lung with a single central nutrient vessel, bronchiolization of the alveolar epithelium, lymphangiectasia and vascular congestion.

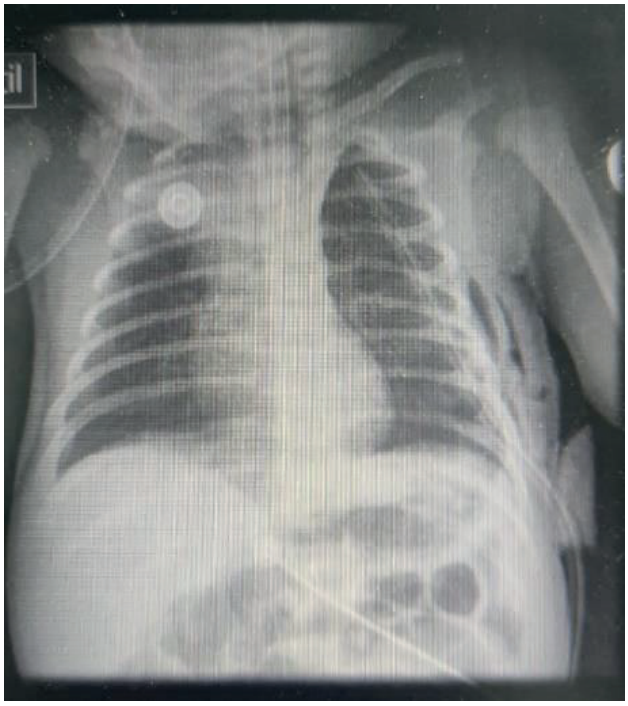


Image 8. IPostoperative radiography.

Discussion

Bronchopulmonary sequestration (BPS) is a rare congenital malformation characterized by a mass of nonfunctional lung tissue that does not communicate with the tracheobronchial tree and receives an abnormal vascular blood supply from the systemic circulation[9].

There is a paucity of epidemiological studies on SBP despite it ranking second as the most common congenital pulmonary malformation, accounting for 0.15–8.3%; 0.42/10,000 live births in China between 2010-2019[9]. Two different theories have been proposed to explain the pathophysiological mechanisms of congenital bronchopulmonary malformations. The environmental hypothesis suggests that a persistent expression of early markers of lung development, caused by potential genetic defects, could lead to a focal and temporal disruption of lung morphogenesis. More recent histological studies of lung malformations have suggested an obstructive hypothesis: focal obstruction of the respiratory tree, whether functional (peristalsis abnormality) or organic (bronchial stenosis), would generate an increase in mediators and cause CPAM abnormalities. Currently, the modes and timing of obstructive events are poorly understood[10].

Stocker et al. They initially identified three stages through macroscopic and microscopic description, which led to the first classification (type 1, the most common form, with large cysts containing mucous cells, type 2 with multiple small cysts and possibly other associated anomalies), type 3 with bulky solid-appearing lesions and frequently displaced mediastinum), which would later be updated by adding type 0 or acinar dysplasia, which is usually fatal, and type 4, which is an acinar malformation characterized by cysts of various sizes. , covered by alveolar cells type 1 and 2 without mucous cells[10].

It generally occurs in early stages of life, although cases have been reported in adulthood where it can manifest acutely and with symptoms such as respiratory difficulty, recurrent pneumonia, frequent infections, presence of purulent sputum, hemoptysis and in cases unusual, hemothorax and heart failure[11].

Polyhydramnios may also occur secondary to compression of the esophagus by the intrathoracic mass and pleural effusion (hydrothorax or fetal chylothorax), which usually resolves when the compression subsides, either due to spontaneous involution, percutaneous chemical sclerosis, or laser sclerosis of the nutrient vessel. Our case progressed from mild polyhydramnios (MBV: 9.1 cm) to severe oligohydramnios (MBV: 1 cm) spontaneously in 4 weeks with resorption of the pleural effusion.

Advances in ultrasound technology and the widespread use of prenatal ultrasound have resulted in increased identification of congenital pulmonary airway malformations during pregnancy. This has led to a better understanding of how some of these anomalies evolve naturally and has opened up the possibility of making preparations for childbirth and post-birth care, and even in severe cases, providing prenatal treatment[12–14]. In the literature related to surgery, pathology, and imaging, various terms are used to describe lung abnormalities present at birth. For this reason Kellenberg et al. In their 2020 study, they concluded and proposed a classification based on the findings of specialized fetal lung MRI can accurately identify, localize and categorize congenital anomalies of the pulmonary digestive tract, with high accuracy and very good agreement between readers[15], however this was not used in our case.

In other studies, excellent agreement was observed in relation to MRI results, even between residents and radiologists with more than 10 years of experience, which shows reliable and comparable values for each finding considered, with the exception of identification of venous drainage. Despite the high diagnostic reliability of MRI for bronchopulmonary sequestration, identifying the venous drainage branch represents the most difficult challenge in diagnosis; However, it was recognized mainly by the most experienced radiologists, who

managed to correctly distinguish between the intralobar and extralobar forms in a higher percentage of cases than the other groups[16].

Some studies show that patients who presented symptoms had an unfavorable outcome (longer stay in the intensive care unit and higher incidence of postoperative complications) compared to patients who did not present symptoms, which raises the question from the prenatal period. The main limitation of most studies is that they are carried out retrospectively and that the number of patients included is generally limited by their dispersion and/or diagnostic limitations or access to subspecialists in maternal-fetal medicine and fetal surgery.

Conclusions

Prenatal diagnosis and treatment studies are very limited in our country largely due to the fact that there are only 2 Fetal Therapy centers in the country's public sector (both in the capital Quito), as well as significant diagnostic limitations in our center. There is at least one study in the literature that compares two therapeutic options in the neonatal period; however, we did not find any national prospective studies in this regard. In our case, the main limitation to achieving the desired goal of 39 weeks and early hospitalization due to therapeutic non-compliance on the part of the patient, with no apparent impact on the outcome of the pregnancy. It is recommended to carry out prospective studies with a more representative sample of patients to determine more clearly which is the best therapeutic option8, as well as to streamline the flow of patients and their early access to medical services subspecialized in fetal medicine.

Patient perspective

The patient was always satisfied with the diagnostic, therapeutic and follow-up plan established and give us all permission to use the information related to her son clinical case. After almost having to end the pregnancy at 33 weeks and keeping the patient hospitalized for the next 4 weeks with her consent and that of her closest relatives, I managed to understand the real life options of her future child, and the possible complications. immediate neonatal measures that included both a satisfactory spontaneous evolution and neonatal surgery since we did not have the option of fetal surgery in the province. She expressed gratitude at every moment of the care and follow-up process and provided consent for the use of all the information in the case, including personal data, spontaneously and in full use of her faculties.

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