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Case Report
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Bilateral Choanal Atresia in a Newborn: a Case Report

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ABSTRACT

Introduction: bilateral choanal atresia is a rare condition that puts the life of the newborn at risk due to an obstruction between the nasal cavity and the oropharynx. Its exact cause is still unclear, it is believed to result from a malformation during embryogenesis between the 4th and 11th gestational weeks, leading to the resorption of the buccopharyngeal membrane and the persistence of Hochstetter's nasobuccal membrane, resulting in obstruction[1].

Methods: we escribe a case of bilateral choanal atresia, aiming to point out to healthcare professionals the importance of early diagnosis of the condition, given its rapid progression to a neonatal emergency.

Results: we describe a case of a pre-term newborn who, after clinical complications, a nasal endoscopy was performed, revealing turbinate hypertrophy, mild septal deviation to the right, total stenosis of the left nasopharynx with secretion, and total stenosis of the right nasopharynx. The surgical correction was successfully performed.

Conclusion: the immediate recognition of cases of choanal atresia is extremely important for the survival of patients. Assertive diagnosis and provisional and definitive treatments reduce the risk of complications.

K**eywords:** Choanal atresia; Surgical treatment; Newborn.

Introduction

Choanal atresia may be related to CHARGE syndrome in 60% of cases, with its main features being coloboma, congenital heart disease, choanal atresia, growth and developmental delays, genital anomalies, and ear anomalies with or without hearing loss, which give the condition its name[1,2,3,4]. Its exact cause is still unclear, it is believed to result from a malformation during embryogenesis between the 4th and 11th gestational weeks, leading to the resorption of the buccopharyngeal membrane and the persistence of Hochstetter's nasobuccal membrane, resulting in obstruction[1,3].

In most cases, choanal atresia is recognized at birth during the initial postnatal care. Nasal probing, followed by computed tomography of the airways, generally confirms the diagnosis[1,5,6]. When not diagnosed at birth, it can progress to cyanosis, noisy breathing, asphyxia, and respiratory failure, becoming a neonatal emergency. Since newborns are obligate nasal breathers until the third week of life- due to the larynx not having descended and the soft palate necessarily resting on the vallecula, preventing the natural patency of the oral airway-they require a patent nasal airway to avoid imminent risk of death [1,5,7].

The description of a case of bilateral choanal atresia aims to emphasize to healthcare professionals the importance of early diagnosis of the condition, given its rapid progression to a neonatal emergency. Additionally, it highlights the positive outcomes achieved through swift and decisive interventions. Based on these principles, an improvement in the quality of life for newborns with bilateral choanal atresia can be ensured.

This study was approved by the Human Research Ethics Committee, under opinion number 6.552.140, and follows the writing guidelines of the CARE Guideline.

Case Report

Preterm newborn, female, small for gestational age. Born to a 28-year-old mother, second pregnancy with planned pregnancies (G2P2A0), blood type A+. During prenatal care, the mother had negative serologies, susceptible to rubella, and susceptible to toxoplasmosis. She was diagnosed with gestational hypertension and was on Methyldopa 500mg since the second month of pregnancy. She had more than ten prenatal consultations, with quarterly ultrasounds indicating fetal growth appropriate for gestational age. There were no complications during the pregnancy. At 36 weeks and 1 day, the mother presented with preeclampsia and preterm labor.

The child was born via cesarean section weighing 2000 g, with Apgar scores of 4 and 7. She was born in apnea and progressed to cardiorespiratory arrest, requiring resuscitation in the delivery room. She immediately received positive pressure ventilation and orotracheal intubation. She was transferred to a nearby tertiary hospital and admitted to the neonatal Intensive Care Unit (ICU) (Figure 1).

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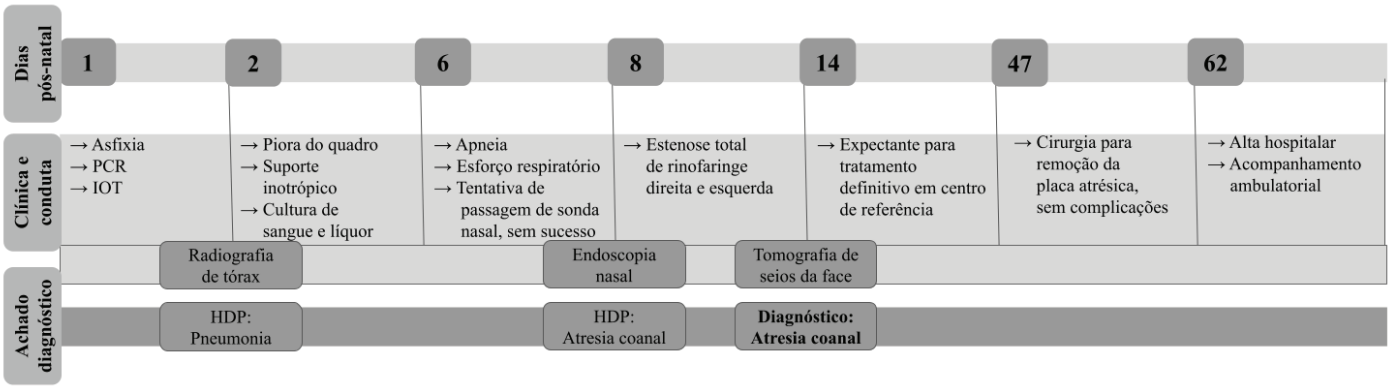

Figure 1. Timeline relating the clinical picture to the patient's postnatal diagnostic exams.

On the first day postpartum, the child experienced a seizure, which was controlled with a loading dose of Phenobarbital at 20 mg/kg and a maintenance dose of 4 mg/kg/day. A chest X-ray revealed opacity in the middle third of the left lung, indicative of pneumonia. A regimen with Ampicillin and Gentamicin was started. While on mechanical ventilation, she received enteral nutrition through a feeding tube.

After 2 days of birth, the child showed worsening clinical progression. She had dehydrated and pale mucous membranes and poor peripheral perfusion, leading to the initiation of inotropic support with Dobutamine and Dopamine. Laboratory tests, including a complete blood count, arterial blood gas, lactate, and transfontanel ultrasound, showed no significant abnormalities except for metabolic acidosis. Results of blood culture and cerebrospinal fluid analysis were pending, and a transfontanel ultrasound was scheduled. During this period, she also developed jaundice and was placed under phototherapy. The child gained weight and maintained stable vital parameters in the following days.

On the 4th day after birth, phototherapy and Dobutamine were discontinued, while Dopamine was continued until the following day. The child remained on mechanical ventilation.

On the 6th day after birth, the newborn continued to have good weight gain and stable vital signs. During a routine physical therapy session, an attempt was made to pass an aspiration tube through the upper airway, but it did not progress through either the right or left nostrils. The baby was extubated and maintained on a 30% oxygen halo, not showing respiratory discomfort at that time.

On the 8th day postpartum, the newborn continued to gain weight and had stable vital signs. During a bath, she experienced an episode of cyanosis, desaturation, and apnea, requiring reintubation and continued mechanical ventilation. On the same day, another attempt was made to pass the aspiration tube through the upper airways, again without success in either nostril. After these two events, an otorhinolaryngologist was consulted for suspected bilateral choanal atresia, which was confirmed on the 10th day of life.

The otorhinolaryngologist made another attempt to pass an aspiration tube through the nostrils, without success. A nasal endoscopy was performed, revealing: turbinate hypertrophy, mild septal deviation to the right, total stenosis of the left nasopharynx with secretion, and total stenosis of the right nasopharynx. With the primary diagnosis confirmed by endoscopy, the otorhinolaryngologist requested a CT scan of the paranasal sinuses.

On the 14th day after birth, the child continued with stable vital signs, on mechanical ventilation, and had good weight gain. The CT scan of the paranasal sinuses showed: significant bilateral narrowing of the posterior airways, indicating bony choanal atresia, with mucous retention in the nasal cavity and air-fluid levels above the point of obstruction; paranasal sinuses appropriate for the age group; centered nasal septum; and preserved pterygopalatine fossae (Figure 2). After confirmation of bilateral choanal atresia, the McGovern nipple adapter (Figure 3) was fitted, and the child was extubated. On the same day, contact was made with the pediatric referral center for the child's transfer and definitive treatment.

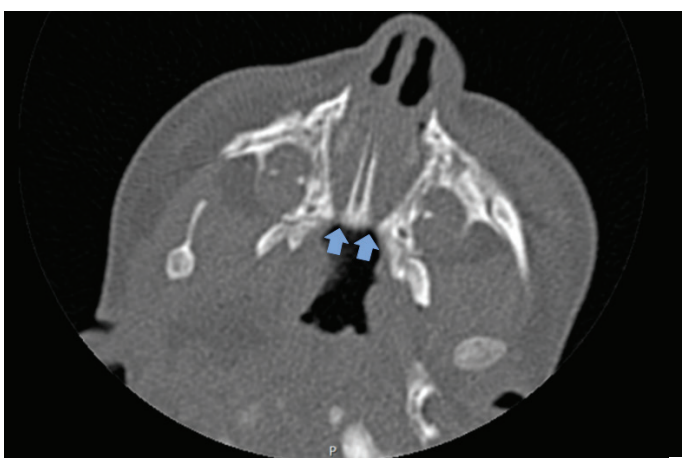

Figure 2. Computed tomography of the facial sinuses with significant bilateral narrowing of the posterior airways, characterizing bone atresia of the choanas (identified by blue arrows). Nasal mucoid retention with air fluid levels seen above the point of obstruction; paranasal sinuses common to the age group; centered nasal septum; and preserved pterygo-palatine fossae.

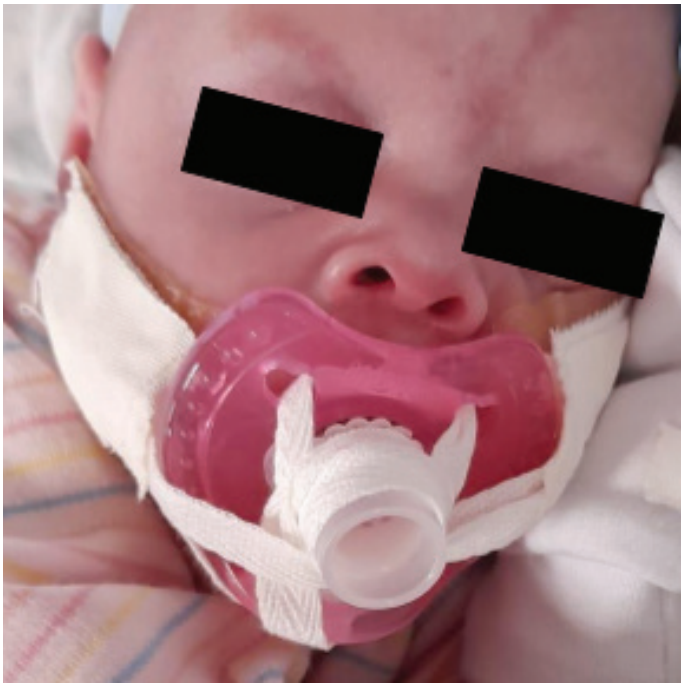

Figure 3. Adapted McGovern pacifier maintaining the patency of the newborn's upper airways.

After contacting the referral center, it was advised to transfer the child only after completing the first month of life, corrected for term birth age, and reaching a weight milestone of 3500 g.

On the 19th day after birth, the child was in good general condition, weighing 2320 g, with good adaptation to the McGovern nipple and breathing in room air. She continued in this condition for the next 13 days. On the 32nd day postpartum, weighing 2815 g, a lesion was noticed on the child's palate, and the McGovern nipple was adjusted to a softer latex, which corrected the lesion. On the 34th day postpartum, the child developed respiratory discomfort, with stable other signs and good weight gain, weighing 2970 g. The McGovern nipple was readjusted with an increase in the length of the nasopharyngeal tube (previously attached in a smaller size), with partial improvement in the condition. A chest X-ray showed a central diaphragmatic eventration, possibly associated with regional atrophy. A thoracic surgeon's evaluation confirmed the suspicion.

On the 36th day after birth, the child's respiratory effort worsened. Ventilatory support was reinstated using a 40% oxygen halo, which improved the condition.

Another contact was made with the referral center for the child's transfer. On that date, the child was exactly one month old by corrected age and weighed 2995 g.

At 46 days after birth, weighing 3400 g, the transfer for the correction of choanal atresia was accepted.

On the 47th day postnatal, already at the referral center, the child underwent bilateral resection of the atresia plate without surgical complications. She was admitted to the ICU for monitoring post-surgery. She remained in the ICU for the next 3 days, using nasal Dexamethasone without complications.

On the 4th day postoperatively, she was transferred to a regular ward, ceased Dexamethasone use, and continued with good clinical progress. During this hospitalization, an investigation for genetic syndromes was conducted due to suspected craniosynostosis. The requested genetic molecular panel would be ready in three months and should be evaluated on an outpatient basis in the city of origin.

After 15 days postoperatively, the child was discharged from the hospital. She remained in excellent general condition throughout the postoperative period, with no respiratory discomfort, saturating 100% in room air, and good feeding acceptance.

At the time of discharge, it had been about 2 months since birth. Instructions were given to the child's mother on how to perform daily nasal washes, and an outpatient follow-up at the referral center was scheduled for two weeks later. The child and mother returned to their hometown.

Two weeks after hospital discharge, at an outpatient visit, the mother reported that the child was well, without respiratory effort, performing nasal washes, with feeding acceptance (breastfeeding and complementary formula), and no complications during the period. During physical examination, the choanae were patent on nasal endoscopy, with good surgical healing. At 8 months and 16 days, the child returned to the referral center's outpatient clinic, with no complications, showing similar progress to the previous visit, and was discharged from outpatient care.

Discussion

Bilateral choanal atresia is a rare condition that puts the life of the newborn at risk due to an obstruction between the nasal cavity and the oropharynx[1,2]. It was described in 1755 by Roederer, disclosed as an anatomical alteration for the first time in 1829 by Otto and the first corrective surgery was performed in 1851 by Emmert. In most cases, bilateral choanal atresia is identified at birth, during the first care given to the newborn, during the verification of nasal patency using a probe[2,3,4]. The unfavorable evolution begins when the diagnosis is late, leading to respiratory failure and possible death[1,3,4].

The pathophysiology of bilateral choanal atresia is related to a gestational malformation, occurring between the 4th and 11th week of embryogenesis. Of the various hypotheses associated with this error, the most accepted are related to the lack of reabsorption of the buccopharyngeal membrane, the persistence of Hochstetter's nasobuccal membrane and the change in the orientation of the mesodermal cells that will constitute the nasal cavity[1,2,3,4,5,6,7,8]. Considering the subtle morphological changes during pregnancy, cases

of bilateral choanal atresia are recognized after birth, as in the present case.

This disease may present congenital clinical manifestations associated with CHARGE Syndrome (such as coloboma, heart defects, growth retardation and mental retardation, genital and ear anomalies)[1,2,13,14]. Furthermore, pre-eclampsia is an additional risk factor for growth retardation[13,14]. Despite bilateral choanal atresia being highly related to genetic malformations and the presence of a risk factor, the newborn in this case did not present congenital manifestations. Craniosynostosis, another congenital alteration little associated with bilateral choanal atresia[14], was investigated in neonates, given changes in the formation of their skull. But without a sequential report. Consequently, the absence of congenital components may delay the investigation of newborns with bilateral choanal atresia.

Additionally, among the clinical manifestations we have nasal respiratory discomfort, generally presented at birth, with rapid progression to cyanosis. Cyanosis in most cases presents itself cyclically, with obstruction during rest and relief when the newborn resumes breathing[1,4]. When the condition is not identified quickly, it can progress to noisy breathing and consequently respiratory failure, generating a neonatal emergency[1,3,4,5]. The newborn studied was born in apnea, progressing to cardiorespiratory arrest. An emergency scenario related to respiratory distress. After the reversal of the cardiac arrest while still in the delivery room, the newborn was transferred to a tertiary hospital, where he received continued care. However, the suspected diagnosis of bilateral choanal atresia was not confirmed, since the assessment of airway patency was not performed.

The diagnosis of bilateral choanal atresia must initially be clinical, based on suspicion based on clinical manifestations. This can be confirmed in two ways, via flexible nasofibroscopie or by computed tomography of the sinuses. The latter has become the gold standard, due to the exact visualization of the location of the atretic plate and its composition, generally osteo mucosa[4,8,9,12]. The case in question was not diagnosed shortly after birth. Only after follow-up and hospitalization of the newborn was an attempt made to insert a nasal aspiration tube. The suspicion of choanal atresia arose after the procedure was not completed in both nostrils. The definitive diagnosis was made using the gold standard method.

The treatment of bilateral choanal atresia must be immediate, as newborns are mandatory nasal breathers until the third week of life, thus avoiding unwanted events[12]. Temporarily, maintaining a patent airway can be done with the use of a McGovern pacifier or Guedel cannula. In some cases where adequate ventilation is not achieved, orotracheal intubation is necessary[8,9,10]. Orotracheal intubation is avoided due to the risk of complications, such as pneumonia associated with mechanical ventilation, for example. The newborn in the case described presented the aforementioned complication after his first intubation, right at birth during cardiorespiratory arrest. Supportive treatment with the McGovern pacifier was performed in this case, with good acceptance by the newborn. Ventilation adaptations during the management of choanal atresia cases are common; in our newborn, a series of adjustments were necessary and successful, guaranteeing the occurrence of major complications.

The definitive treatment of choanal atresia is performed by surgical correction, generally after a few hours of life[8,9,10,11]. The literature differs on the safest surgical method. The main complication is the occurrence of choanal restenosis. In these cases, stent implantation during surgery is subject to discussion[9,10,11,12]. A study reviewing medical records[10] showed that among 18 cases, transpalatal surgery followed by stent placement was recommended. In only one of the cases where the approach was endoscopic and without stent placement, there was restenosis. Thus, most current authors agree with transpalatal or endoscopic surgical correction with stent implantation[10,11]. The service where the newborn in this case was diagnosed does not provide definitive treatment for choanal atresia, resulting in a long hospitalization while waiting for transfer to the referral hospital.

The average length of hospital stay after definitive correction of bilateral choanal atresia is 5 days, with outpatient follow-up after this period. During outpatient follow-up, nasal endoscopy is recommended in order to identify cases of restenosis early[12,15]. After removal of the atretic plate, the newborn in this report remained hospitalized for 15 days, without complications during the period. The control endoscopic examination was satisfactory.

This study has limitations related to the lack of specific data recorded in the medical records, such as dosage of medications used and description of the surgical approach, without information whether a stent was implanted. Furthermore, the result of the craniosynostosis investigation could not be accessed.

The strengths of the study are mainly related to the detailed description of the case, from birth to outpatient follow-up of the newborn. Furthermore, this report reinforces the need for early recognition of cases of choanal atresia, in addition to competence in supportive and definitive treatment. Anticipating such conduct avoids prolonged hospitalizations and risk to the newborn. In this sense, patients with bilateral choanal atresia benefit from being treated in a center with experience and qualified staff.

Conclusion

The recognition of cases of choanal atresia, given the clinical manifestations and the possibility of

rapid evolution, is extremely important for the survival of patients. Assertive diagnosis and provisional and definitive treatments reduce the risk of complications. Outpatient monitoring by the team is essential for the patient's complete recovery. In this case of bilateral choanal atresia, therapeutic management was optimized soon after diagnosis, with positive repercussions and an excellent outcome for the patient and his family.

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