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CASE REPORT

Deglutition disorder in children with interstitial lung disease complex: therapeutic assessment and proposal

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Abstract

Case report of a 7 month old baby with the disease studied, in which the evaluation and therapeutic proposal of the swallowing disorder was performed in home care. In the speech-language evaluation hypotonia of the musculature of the stomatognathic system was identified, and bite and GAG reflexes were present. It presented little tolerance to the intra-oral touch, with signs of stress during evaluation. A direct evaluation of swallowing was performed, offering 5ml of food in a homogeneous pasty consistency in which we observed respiratory incoordination and slow oral transit time, with food stasis in the oral cavity. Patient presented respiratory discomfort, crying, and irritability, with the presence of regurgitation of mucoid secretion and the refusal to a new attempt of supply. Patient with speech-language diagnosis of oropharyngeal dysphagia, with signs of eating difficulties. Speech-language intervention, global sensory stimulation, guidelines for those responsible and the nursing techniques team on posture, and therapeutic and stimulation activities were conducted. Patient presents a rare complex of pulmonary diseases and speech-language pathology is innovative in patients with this pathology.

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INTRODUCTION

Interstitial lung diseases (ILD) constitute a heterogeneous group of disorders, difficult to diagnose due to changes to the lung walls and alveolar spaces, and it can also be called diffuse parenchymal lung disease[1]. The presence of ILDs in the pediatric group suggests a prevalence of 3.6 per million[2]; however, it is not truly known; in most cases it is underdiagnosed and underreported, but rare in children[3].

Eating problems in these children with respiratory disorders have been studied; an association of swallowing disorders in infants with lung diseases has already been found, showing the interference of the respiratory condition in the swallowing process[4]. Given the rarity of this disorder, and the lack of descriptions of clinical data on swallowing in patients with ILD, this paper aims to describe the assessment and treatment of swallowing in a patient with ILD.

CLINICAL CASE PRESENTATION

Case study, male patient, treated in the city of São Leopoldo/RS, after the guardians signed the Informed Consent Form. The present study was developed following the precepts of Ethics in Scientific Research in accordance with resolution 466/12, under embodied opinion nº: 2.492.475.

Patient information was obtained through anamnesis with the patient's guardian, assessment of structures of the stomatognathic system and clinical assessment of swallowing.

Birth information: Mother of 37 years old, in the second pregnancy, the first being uneventful. Normal delivery, 39 weeks gestational age, baby weighing 3,320 grams and 52 centimeters long. Apgar 9/9, with the presence of early respiratory dysfunction, requiring admission to a neonatal intensive care unit due to respiratory dysfunction resulting from pneumomediastinum.

Postpartum info: Oxygen was used through a cannula. He was submitted to radiological control until disappearance of the pneumomediastinum, maintaining the interstitial pulmonary infiltration. Chest tomography showed bilateral interstitial infiltration, with greater intensity on the right side, with a ground-glass appearance (Figure 1). He remained hospitalized for 217 days, during which he developed respiratory distress, and required oxygen therapy. He was under a chest tube and mechanical ventilation for 1 week; and later, he was submitted to noninvasive ventilation and a high-flow catheter, until he was discharged with a tracheostomy. Alveolar proteinosis, interstitial pulmonary glycogenosis, pulmonary hypertension, alveolar underdevelopment, and surfactant protein disorder was clinically diagnosed via pulmonary biopsy. He remained with a naso-gastric tube until 5 months of age, when a gastrostomy was performed.

During hospitalization, at 6 months of age, the patient underwent speech therapy evaluation; however, the mother reported that the patient did not accept the intervention, due to constant episodes of crying and consequent cyanosis. He

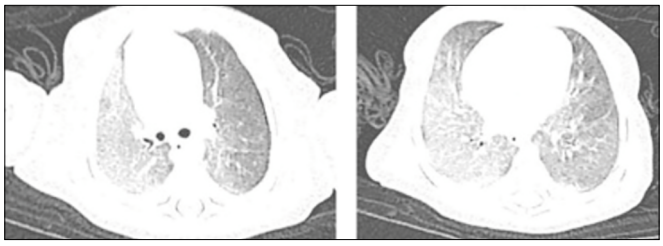


Figure 1. Chest CT showing bilateral interstitial infiltration.

was submitted to speech therapy 2 to 3 times a week, for overall sensory and tactile stimulation. The Blue Dye Test (saliva dye with 4 drops of blue food coloring and subsequent tracheostomy aspiration, and had a colored secretion oozing out) was performed by the speech therapist of the hospital, with a negative result, indicating the absence of laryngotracheal aspiration. Deprived of oral feeding from birth and no acceptance of stimuli during hospitalization, even after monthly attempts.

Upon the home assessment, a 7-month-old patient, with a diet via gastrostomy, using mechanical ventilation at controlled pressure in the tracheostomy (Parameters: PIP: 17.1 cm H2O, maximum flow: 17.2 liters per minute), in a deflated cuff. The speech-therapy assessment was performed with the patient seated and neck support, the respiratory rate was 31 breaths per minute, heart rate of 103 beats per minute, and peripheral oxygen saturation of 95%. The patient was alert, with interaction with the examiner, no signs suggestive of respiratory distress. The following altered items were observed in the structural assessment: lips and cheeks with reduced tone, tongue at rest in the mouth floor with reduced tone. Presence of protective bite and GAG reflexes. Hyperreactivity and hypersensitivity to extra and intraoral touch, with the presence of nausea on contact with the tongue. In the assessment of salivary swallowing, the patient had an adequate appearance of the oral mucosa and swallowing frequency within the normal range, without signs suggestive of secretion penetration and/or laryngotracheal aspiration.

Direct evaluation of swallowing was carried out, with an offer of 5 ml of food in a homogeneous pasty consistency (carrot cream), colored (insertion of 4 drops of food coloring in the food and offering it to the patient, seeing the blue coloring via tracheostomy, or not). He had inadequate food content uptake, offered with a silicone spoon, and a bite reflex. He had proper tongue movement, with a frequency of 2 swallows per food portion, with respiratory incoordination and slow oral transit time, with food stasis in the oral cavity. The patient had a reduction in the hyolaryngeal complex elevation, which was restricted due to the tracheostomy. The patient presented respiratory distress, crying, and irritability, with the presence of regurgitation of mucoid secretion and refusal to make another attempt. Tracheal aspiration was performed after oral administration, with no blue color output, with negative result of the Modified Blue Dye Test.

After swallowing clinical evaluation, the patient presented a diagnosis of Moderate to Severe Oropharyngeal Dysphagia. However, in addition to the oropharyngeal dysphagia, there were signs of eating difficulties, possibly related to the association of some factors, such as the evolution of the underlying disease, history of prolonged hospitalization and deprivation of oral feeding since birth.

The home speech therapy for this case was established with the frequency of three weekly sessions, of one hour each. Initially, the therapy was aimed at raising awareness among parents and staff in general about the patient's diagnostic hypothesis. The patient's guardians were given extra-therapy sensory stimulation instructions, with manipulation of objects of different textures and temperatures in the body extremities. The parents were instructed on the need to experience moments of family feeding, associating it with a pleasurable action, when it was agreed that the patient would be placed in a feeding chair during the parents' meals.

The speech therapist, in addition to the guidelines, instructed on the overall sensory stimulation with food of different textures, different temperatures and vibratory stimuli. Also, different foods were presented for exploration and knowledge, allowing the patient to manipulate and explore them with any utensil and any part of the body that he felt comfortable with (Figure 2).

The patient has progressed in the overall development, exploratory behavior when exposed to new textures and foods; however, the interest is not maintained for more than 20 minutes, requiring new approaches in the remainder of the treatment session. Family support is crucial for the success of the intervention. The parents participated actively in all stages of the treatment, understanding the patient's underlying pathology, and were instructed on the approach to better help the patient in such condition.

COMMENTS

The diagnosis of several pathologies of the respiratory function is directly linked to the oropharyngeal dysphagia found in this case. This patient had an alteration in the respiratory function, concomitant with the need to use a tracheostomy. This, in turn, caused changes in myofunctional aspects such as sensory loss, changes in smell and taste, leading to changes in the anticipatory phase of swallowing; where there will be a reduction in appetite. Another physiological impact resulting from the tracheostomy is the change in the temperature lower airways flow, which provides desensitization of the mucosa with a risk of silent aspiration[5]. Also, the use of mechanical ventilation causes changes to the swallowing physiology, with changes in tongue and cheek tone, desensitization of oropharyngeal structures, compromising laryngeal elevation and swallowing-breathing coordination[6].

Feeding is a complex, learned, and improved process that depends on the child's physical status and development,



Figure 2. Therapy for feeding refusal.

as well as their oral and sensory abilities to interpret and deal with food. This complex function involves emotions, motivation, social and family context and environment[7]. A study reports that 30% of children with normal development may have some challenges in this process[8], and it is estimated that 80% of eating disorders are found in those with developmental alterations[9].

There is a biopsychosocial model to approach and understand the treatment of children with eating difficulties, which considers physiological, behavioral, and social factors. It is the so-called understanding the "whole child", encompassing the sensory, motor, oral motor, behavioral, learning, physical and nutritional, emotional, and environmental areas. These areas have the same importance and should be connected for the diagnosis and rehabilitation of children with food aversion[10,11].

For the treatment of food aversion in this case, we followed the description by Junqueira (2017)[7], which lists six essential steps in the rehabilitation of these children with eating difficulties, namely: identifying and treating possible organic causes; educating parents; emotional support to the mother; normal sequence development for learning to eat; ensure skill and comfort in feeding; playful strategies. Eating skills are taught and learned. The patients can see, interact, smell, and taste food. The child knows and relates to the world

through play. Therefore, this entire process has been based on playful activities related to food.

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