Intestinal pseudo-obstruction: A rare presentation of congenital hypothyroidism

Pseudo-obstrucción intestinal: una presentación poco frecuente de hipotiroidismo congénito

Children with thyroid gland diseases can present with gastrointestinal symptoms, mainly due to alterations in gastrointestinal motility.

A 5-month-old female infant was admitted to the emergency department, presenting with oral feeding intolerance and abdominal distension of 24-h progression. She was from a rural community in Mexico, with young, non-consanguineous parents. Metabolic screening of six elements was carried out, in which, according to the parents, a non-specified alteration was identified. However, due to the SARS-CoV-2 pandemic, the follow-up consultation was postponed. The infant passed meconium within the first 24 h but had a history of constipation during her first two weeks of extrauterine life, presenting with voluminous stools once a week. She also presented with jaundice of undetermined etiology for the first two months.

Upon hospital admission, the infant had severe overall neurodevelopmental delay. She presented with hoarse cry, coarse facies, swollen eyelids and lips, macroglossia, and inability to support her head. She had a restrictive thoracic pattern due to abdominal distension, but no heart murmur upon auscultation. The abdomen was tympanic to percussion and painful when palpated. There were signs of an umbilical hernia and no hepatosplenomegaly. She presented with hypotonic and hypotrophic extremities, with diminished pulses, cutis marmorata, and generalized xerosis.

A plain abdominal x-ray was ordered, revealing important intestinal segment dilation (Fig. 1A). An orogastric tube was placed, with abundant output of bile, and a transrectal tube improved the abdominal distension (Fig. 1B). The infant had clinical signs of low cardiac output that responded to dobutamine infusion and presented with oliguria and elevated serum creatinine and blood urea nitrogen. Thyroid panel results were TSH 404 mIU/mL, total T4 0.420 ng/dl, T4 0.058 ng/dl, total T3 0.195 ng/dl, T3 0.321 pg/mL, for which treatment with levothyroxine was indicated. Feeding was started with extensively hydrolyzed formula in continuous infusion one week after her admission. Given the clinical presentation consistent with Hirschsprung’s disease, a contrast-enhanced barium enema was ordered that revealed an apparent transition zone in the rectosigmoid colon (Fig. 2), and so rectal irrigations were started. A rectal...
biopsy was taken, and high-resolution anorectal manometry was performed, documenting the rectoanal inhibitory reflex. Pericardial effusion, anemia, elevated direct bilirubin, and elevated transaminases were detected in the complementary evaluations.

The patient was released 4 weeks after admission, with improved general clinical status, adequate oral feeding tolerance, and no treatment with laxatives or prokinetics. At the outpatient follow-up, the infant presented with no upper gastrointestinal symptoms or abdominal distension. Her bowel movement pattern was normal, neurologic development showed improvement, and thyroid profile was normal.

The gastrointestinal alterations and symptoms that coexist with congenital hypothyroidism are dysphagia, vomiting, reduced gastric emptying, and small intestinal bacterial overgrowth; intestinal pseudo-obstruction is rare. However, in the directed interview with the parents, the infant did not present with upper gastrointestinal symptoms, until her admission to the emergency department. In the review of the literature, we found only a few reports of intestinal occlusion as a presentation of hypothyroidism in adults. Current recommendations in the approach to pediatric chronic intestinal pseudo-obstruction suggest performing thyroid function tests, mainly when there are signs of hypothyroidism.

The coexistence of hypothyroidism and Hirschsprung’s disease has been reported. Given that the thyroid hormones are essential for histogenesis, cell migration, and the cytoarchitecture of the central nervous system, the lack of those hormones has been suggested to favor the development of Hirschsprung’s disease, due to effects on gene expressions, ligands, receptors, apoptotic factors, or extracellular matrix proteins that affect cell migration or differentiation. Thus, we had to rule out Hirschsprung’s disease in our patient, which we did, through rectal biopsy and anorectal manometry.

In conclusion, the evaluation of congenital hypothyroidism should be considered in patients that present with intestinal pseudo-obstruction. In addition, other entities whose clinical presentations are similar to that of Hirschsprung’s disease must be ruled out. In turn, Hirschsprung’s disease can be related to congenital hypothyroidism.

Ethical considerations

The authors obtained informed consent from the parents of the patient referred to in this article and the document is in the possession of the corresponding author. The present work meets the current bioethical research regulations and because of the nature of this scientific letter, does not require approval by the hospital ethics committee. The authors declare that the article contains no personal information that could identify the patient.

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Conflict of interest

The authors declare that there is no conflict of interest.

References


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