

Gluten-induced Distal Tubular Acidosis?

Nada Boutrid^{1,2}, Hakim Rahmoune^{1,2,*}, Mounira Amrane^{2,3}, Reda Belbouab⁴, Belkacem Bioud^{1,2}

¹Department of Pediatrics, University Hospital of Setif, Setif-1 University, Algeria

²Genetic, Cardiovascular & Nutritional Diseases Laboratory, Setif-1 University, Algeria

³Central Laboratory, CAC Hospital of Setif; Setif-1 University, Algeria

⁴Department of Pediatrics, Mustapha Bacha University Hospital, Algiers, Algiers-1 University, Algeria

*Corresponding author: rahmounehakim@gmail.com

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Abstract The expanding spectrum of gluten-related autoimmune disorders may encompass distal tubular acidosis. We report a case of an infant presenting a classical digestive celiac disease along with a transient, reversible tubular acidosis that vanished after gluten free diet.

Keywords: celiac disease autoimmunity, distal tubular acidosis, gluten

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1. Introduction

Among the rare causes of secondary distal tubular acidosis, we report a case whose panel of signs proved to be completely reversible as soon as the probable immunological cause (i.e. gluten autoimmunity) was treated.

2. Case Description

A 19 months old girl, first child of non-consanguineous parents, is admitted for chronic diarrhea since several months with patent failure to thrive; while the mother also reports a persistent polyuria

On clinical examination, the baby is well hydrated, with mild pallor and sparse hair. She passes 3-4 soft stools a day, without mucus nor blood; and 6-10 micturations in 24 hours. She weighs 8kg (<-2 standard deviations, SD) with a birth weight = 3 kg 200, her height is 83 cm (<-2 SD).

Urine Chemistry reveals pH = 7.5 and Density \geq 1030.

Arterial blood gas confirms metabolic acidosis (pH=7.23).

The blood investigations detect moderate ionic disorders with a natremia = 127 mEq / l, a serum potassium = 2.7 mEq / l and a calcemia = 88mg / l. Kidney and liver function tests were normal, as well as blood levels of glucose, phosphorus and alkaline phosphatase.

The urinary ionogram confirms the renal sodium loss with a natriuria > 30 mmol / l.

X-ray of left wrist denotes osteopenia and bone rickets.

Celiac disease was highly considered and serology found IgA Anti tissue transglutaminase type II = 96 iu.

Upper digestive endoscopy revealed a mosaic appearance of the duodenum mucosa; whereas pathology revealed

villous atrophy, March III (with intra-epithelial lymphocytes count \approx 25%).

After correcting electrolytes and starting gluten-free diet, the patient's appetite improved within a week she passed 2 stools / day with 3-4 urine / day; urinary chemistry noted the disappearance of acidosis (urinary pH= 5, Density = 1005).

After 15 days, her weight gain was 300 g, blood electrolytes were within normal limits, and urine analysis was free of abnormalities.

3. Discussion

According to our knowledge, this is the first ever reported case of transient distal tubular acidosis probably due to celiac autoimmunity.

Literature search on Medline® via Pubmed®: "Glutens"[Mesh] AND "Celiac Disease"[Mesh] AND "Acidosis, Renal Tubular"[Mesh], retrieved no result [1].

Another search on Google Scholar®: allintitle:"celiac OR coeliac" AND "tubular OR renal" AND acidosis found only two cases in patients already suffering from Sjögren's syndrome [2,3] and another case in a teenager with hypothyroidism [4].

In fact, celiac disease is more and more associated to a spectrum of extra-intestinal disorders [5,6]. The peculiar celiac autoimmunity disappears when the triggering agent (gluten) is completely excluded from the patient's diet; allowing resolution of the immunological cascade and its histological and clinical related symptoms.

In the other hand, distal tubular acidosis is considered as a group of disorders of the distal renal tubules characterized by elevated plasma chloride, hyperchloremic metabolic acidosis. Low renal acid excretion at distal renal tubules can lead to complications such as hypokalemia, hypercalciuria with nephrocalcinosis and rickets [7].

Inherited form is due to autosomal recessive mutations in genes encoding subunits of the vacuolar H⁺ATPase, with subsequent transporter function impairment in the renal tubule [8]. Autoimmune, acquired forms are more common in adults, and are often associated with other autoimmune conditions like diabetes or Sjögren's syndrome [9,10].

The quick resolution of biological and clinical signs after introducing the gluten free diet confirms the initial diagnosis; keeping in mind the possibility of a wide autoimmune background.

Close observation and follow up of such forms of celiac autoimmunity is mandatory, especially in a girl with an early-onset burden of disease.

4. Conclusion

The association of isolated celiac disease with extra-digestive signs of distal tubular acidosis brings forth a multiple pathogenesis, probably autoimmune, genetic (HLA) and environmental.

The close follow-up of such cases makes it possible to be on the lookout for another autoimmune attack outside the gluten spectrum; in particular that of a possible Sjögren-Gougerot syndrome.

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