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An adult case of eosinophilic pyloric stenosis maintained on remission with oral budesonide

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KEY WORDS

Eosinophilic gastroenteritis, pyloric stenosis, oral budesonide

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SUMMARY

We describe an isolated eosinophilic pyloric stenosis in a young female. She was referred for abdominal pain, fever, weight loss and eosinophilia. A sonographic examination revealed a concentric pyloric stenosis, with antral palsy and ascites. The endoscopy confirmed the diagnosis of eosinophilic infiltration of the pylorus. After a short course of systemic steroids, the patient was switched to oral budesonide, which effectively maintained a long-term remission. Eosinophilic gastroenteritis limited to pylorus is exceptional in adults, and in our patient it was not associated with allergic other disorders. This case emphasizes the usefulness of sonography for diagnosis and monitoring, and the clinical efficacy of oral budesonide.

The term "eosinophilic gastroenteritis" (EG) encompasses a broad spectrum of diseases, partially related to atopy, and characterized by the eosinophilic infiltration of one or more portions of the gastrointestinal tract (1). EG can be either primary or secondary to infections, parasitic infestations, malignant diseases or haematological disorders, and can be associated with blood eosinophilia (1, 2). In adults the most common form is eosinophilic oesophagitis, whereas the involvement of stomach and duodenum is quite rare. We describe herein a case of eosinophilic pyloric stenosis, primarily diagnosed by ultrasonography, and successfully treated with oral budesonide.

A 23-year old woman was seen at the emergency unit in January 2010 because of persistent vomiting and abdominal pain. Those symptoms lasted about one month, but they suddenly had worsened in the last few days. The past clinical history of the patient was not relevant, with the exception of a 3-kg weight loss in the last month. She was a

non-smoker and denied any recent use of drugs or the ingestion of unusual food. The patient was not febrile, and had only a tenderness in the upper abdominal quadrants, without signs of acute abdomen. The routine laboratory analyses were normal, with the exception of a moderate leukocytosis (16,200 WBC/ μ L) with eosinophilia (25.2%). The erythrocyte sedimentation rate and the C-reactive protein were normal. The abdomen X-ray was negative, whereas the chest radiogram displayed right pleural effusion. An abdominal ultrasonography was immediately performed, evidencing the presence of moderate ascites and of a concentric stenosis of the pylorus, which multi-layer structure was not appreciable. There was no peristaltic movement in the stomach antrum (Figure 1). A sonographically guided paracentesis was carried out, and the microscopic analysis of the effusion evidenced the presence of numerous eosinophils. The gastroscopic examination confirmed the pyloric stenosis. Multiple biopsies were tak-

en, which revealed an intense eosinophilic infiltration of the mucosa. Biopsies taken from oesophagus, stomach and colon revealed a normal structure. The abdominal CT scan, in addition to ascites and pyloric stenosis, identified also some small mesenteric lymph-nodes, but liver, pancreas gallbladder and duct were normal. The search for parasites and ova was negative on several occasions, and total IgE was 231 kU/L. Specific IgE assay for foods and inhalant allergens were completely negative, as well as skin prick tests. Anticytoplasmic, anti gliadin and anti-nucleus antibodies were negative. The diagnosis of eosinophilic duodenitis was established and a systemic treatment with methylprednisolone (80 mg/day) was started. Within few days, all symptoms improved, and the patient could eat normally. The blood eosinophilia disappeared as well as the leukocytosis. The systemic steroid was gradually tapered and replaced with oral budesonide in capsules (Entocir®, AstraZeneca) at the dose of 3 mg twice daily. At a subsequent sonographic evaluation, fifteen days after admission, there was no evidence of ascites, the pylorus had a normal appearance, and the peristaltic movements were present. At present, six months after the acute episode, the patient is still on oral budesonide. She is well and the laboratory analyses remain within the normal range.

Eosinophilic gastroenteropathies are a heterogeneous and still poorly understood group of disorders, characterized by the eosinophilic infiltration of at least one layer of the gastrointestinal wall. (1, 4). A relationship with atopy and/or food allergy has been consistently shown, at least for oe-

sophagitis, but these disorders are not always IgE-mediated. In our patient, in fact, there was no evidence at all of atopy or allergic diseases. Noteworthy, an isolated pyloric stenosis is exceptional in the adulthood, whereas some cases have been reported in children (5, 6). Although a surgical biopsy was not done, we hypothesize that in our case eosinophils infiltrated the whole pyloric wall, reaching the serosal layer. This is supported by the presence of ascites, which is a distinctive feature of serosal involvement. Another remarkable aspect is the role of ultrasonography, which allowed to immediately identify the pyloric stenosis. Although a bioptic procedure was needed to define the diagnosis, ultrasonography could be used for the follow-up controls, so that repeated endoscopic examinations could be avoided. Finally, as previously reported (7, 8), oral budesonide (an inhaled corticosteroid) was effective in maintaining a long-term remission. The special advantage of this treatment is that budesonide acts selectively at mucosal level and has a low bioavailability. This allows to avoid the possible side effects of the chronic use of systemic steroids.

Acknowledgment

We thank Drs C. Spedini and D. Belli for their valuable support in the clinical management of the patient

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Figure 1 - Transversal section of the gastric antrum showing a concentric stenosis (1.5 cm length) at the gastro-duodenal junction. The ultrasound multilayer structure was not appreciable. No peristaltic movement or transpyloric fluid flow were present

