

Esophageal candidiasis in an immunocompetent girl with chronic anaemia.

Barros García Patricia*, Izquierdo Martín Ana, De Nicolás Jiménez Jorge Manuel, López Lafuente Amparo, Mateos Rodríguez José María, González Núñez M Ángeles, López-Rodríguez M José

Department of Pediatrics, Hospital San Pedro de Alcántara, Cáceres, Spain

Accepted on September 02, 2016

Introduction

Infection is an uncommon cause of esophagitis among the paediatric population. The main risk factor is immunodeficiency. In children immunocompromised due HIV infection, certain endocrine diseases, and corticosteroid or immunosuppressive therapy, *Candida albicans* is the most common etiologic agent. The prevalence of *Candida* esophagitis has increased because of an improvement in survival and management of immunological diseases. Even though it is not frequent in immunocompetent children, there have been reported cases related with risk factors [1] such as antibiotic intake (which allow bacterial overgrowth and fungal colonization), proton pump inhibitors (which produce hypochlorhydria allowing oral bacteria to colonise gastric mucosa, and to increase the risk of infectious esophagitis), esophageal obstruction (aperistalsis and stasis helps the fungal overgrowth) and systemic or inhaled corticotherapy (that suppresses lymphocyte and granulocyte function) [2]. *Candida* esophagitis has also been reported in healthy patients, with no evidence of causal mechanism [3]. The most common symptomatology consists in odynophagia, dysphagia and retrosternal chest pain, but epigastralgia, vomiting and gastrointestinal bleeding can also occur. Chronic or acute anaemia could be the sole sign of an asymptomatic presentation [4,5]. The absence of oral candidiasis does not exclude the diagnosis of *Candida* esophagitis [4]. The pathophysiology of the infection is complicated. It includes a large number of factors, such as fungal virulence, mucosal integrity and host susceptibility. Esophagoscopy is the most specific and sensitive method of diagnosis. Characteristic endoscopic findings are confluent or isolated white plaques adhered to the mucosa with edema, erythema and friability, with or without ulcers covered by exudate. The diagnostic confirmation is performed by demonstrating the presence of pseudohyphae or by obtaining evidence of tissue invasion on mucosal biopsy [6]. Fungal culture does not allow differentiation of colonization from infection [4]. The treatment of choice is fluconazole, but there are new therapeutic options [6].

Case Report

A 9-years-old afebrile girl was admitted to our hospital for an episode of witnessed loss of consciousness, increased respiratory secretions, facial cyanosis, without tonic-clonic movements, which lasted a few seconds. She was born at 37 weeks gestation, through an emergent cesarean section due to severe perinatal asphyxia. At the age of 2 and 18 months, she was admitted to the hospital for pulmonary aspiration. Diagnosed with cerebral palsy (CP), psychomotor retardation, refractory epilepsy and central precocious puberty. Treatments included continuous topiramate, levetiracetam and omeprazole, for several years. The patient had not required antibiotics for an extended period and could be fed orally only with a soft diet, because of an oropharyngeal

dysphagia. She had no history of vomiting, dysphagia, abdominal pain, regurgitation or choking. The stools were normal. Physical examination: Weight: 40 kg (97% percentile). Height: 142cm (90% percentile). T: 36,9°C. HR: 84 bpm. RR: 20 rpm. Sat O₂: 92%. BP: 99/53mmHg. Hemodynamically stable. No signs of respiratory distress. Pale skin and mucous membranes. Well hydrated. Hyperemic oropharynx with white friable plaque, easily bleeding to pressure. Pulmonary auscultation significant for loud coarse upper airway sounds. Spastic tetraparesis. Laboratory studies showed: hemoglobin 6.2g/dl, hematocrit 21.8%, mean corpuscular volume 52.7fl, leukocyte count, platelets and coagulation test within normal range. Serum glucose, electrolytes, renal profiles, albumin, total protein, transaminases normal. Serum iron 14 µg/dl, ferritin 4 ng/ml. IgA antiendomysial and IgA antitransglutaminase test were negatives. Humoral and cellular immunologic study normal. HIV negative. Oropharyngeal exudates culture: *C. albicans* isolated. Negative stool hemocult. Chest X-ray normal. Gastrointestinal upper endoscopy: several white patches throughout the distal esophagus, with an edematous friable mucosa (**Figure1**). Esophageal biopsy: focal signs of acute inflammation and presence of pseudohyphae. Findings consistent with *Candida* esophagitis (**Figure 2 and 3**). 24 hours esophageal-pH-metry: no able to assess due patient intolerance. The patient received a blood transfusion and supplemental iron, with an improvement in hemoglobin. She was started on therapy with fluconazole 5 mg/kg/day for 3 weeks. Repeat esophagoscopy was performed two months after, which demonstrated complete resolution of the lesions. Control blood count was normal after three months on iron therapy.

Discussion

Esophageal candidiasis is a rare entity in the paediatric population. Immunocompromised patients are the main risk group. Here we report the case of an immunocompetent paediatric patient with *Candida* esophagitis, but with other potential risk factors, like the treatment with proton pump inhibitors for a long period of time [1,2,5]. There are defense mechanisms that maintain normal flora and inhibit fungus colonization such as salivation, esophageal clearance and mucosal integrity. It has been described that proton pump inhibitors produce hypochlorhydria, which induces changes in gastric bacterial flora and allows infection.

The deglutition process requires a high level of coordination between brain and the enteric nervous system. Patients with cerebral palsy usually have deglutition pathology due to motor disorders, which affect salivary flow [6]. There are limited data regarding salivary production in CP patients, but studies, like Senner et al [7], describe decreased salivary flow, which could be related to the motor disorders responsible for less frequent deglutition. All of these factors can potentially mitigate the

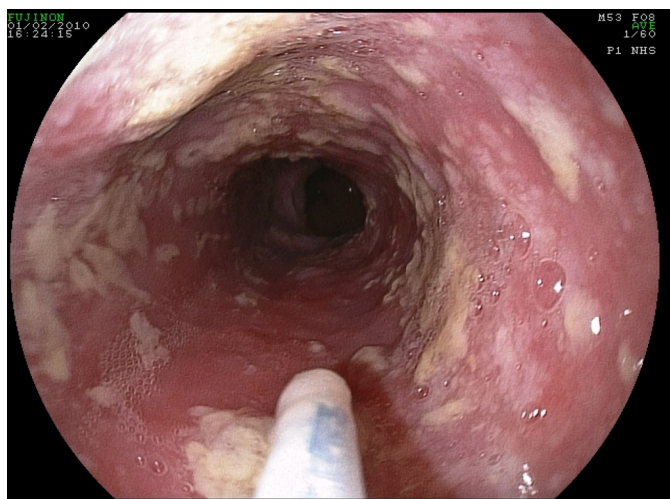


Figure 1: Gastrointestinal upper endoscopy: several white patches throughout the distal esophagus, with an edematous friable mucosa.

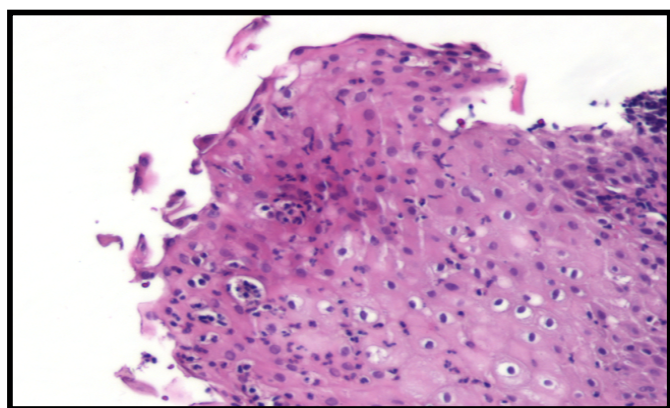


Figure 2: Esophageal biopsy (hematoxylin and eosin stain): focal signs of acute inflammation.

defense mechanisms against *Candida* colonization. Therefore, there were risk factors in our patient, which could have facilitated *Candida* infection.

Gastroesophageal reflux due to brain injury occurs in up to 50% in CP patients [6]. Our patient was on treatment with omeprazole to prevent potential gastroesophageal reflux disease. We were not able to perform a 24 hours esophageal-pH-metry, but histologic results rule out reflux esophagitis.

Although the typical clinical manifestation includes digestive symptoms [8,9,10], there are cases reported without digestive complaints, as in our patient, where anemia was the only indication for the endoscopy. A study realized by Ortuño et al. [8], described a group of immunocompetent adults with *Candida* esophagitis. 29% of the patient had anaemia, which was the endoscopy indication.

In Summary

We describe a new case of *Candida* esophagitis in a paediatric immunocompetent patient. She had associated risk factors such as proton pump inhibitors intake and cerebral palsy, which could favor fungal colonization and the infection.

Reference

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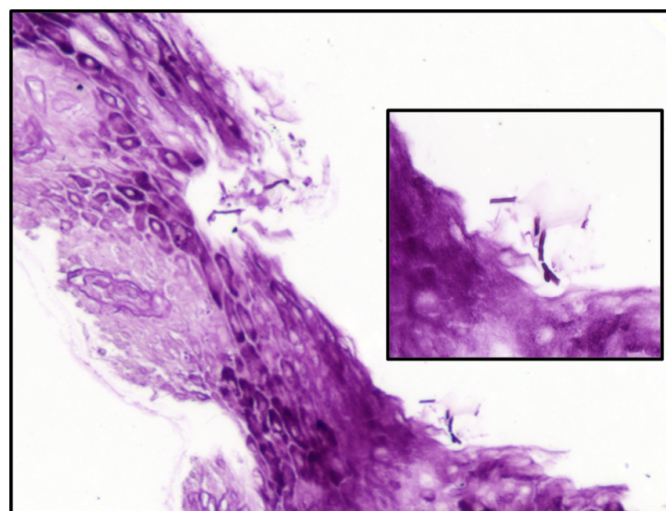


Figure 3: PAS staining of esophageal biopsy showed pseudohyphae.

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*Correspondence to:

Patricia Barros García
Department of Pediatrics. Hospital San Pedro de Alcántara. Avda
Spain
Tel: 0034 927256200
Fax number: 0034 927256202
E-mail address: pbarrosg@yahoo.es