GE Port J Gastroenterol. 2016;23(5):259–263

Portuguese Journal of Gastroenterology

www.elsevier.pt/ge

CLINICAL CASE

Tracheitis – A Rare Extra-Intestinal Manifestation of Ulcerative Colitis in Children

Isabel Serra Nunes[a,*], Marlene Abreu[a], Susana Corujeira[a], Juliana Oliveira[a], Marta Tavares[a], Cristina Rocha[a], Joanne Lopes[b], Fátima Carneiro[b,c,d], Jorge Amil Dias[a], Eunice Trindade[a]

[a] *Pediatric Gastroenterology Unit, Centro Hospitalar São João, Porto, Portugal*
[b] *Pathology Department, Centro Hospitalar de São João, Porto, Portugal*
[c] *Medical Faculty of the University of Porto, Porto, Portugal*
[d] *IPATIMUP – Institute of Molecular Pathology and Immunology of the University of Porto and i3S – Instituto de Investigação e Inovação em Saúde, Universidade do Porto, Porto, Portugal*

Received 18 January 2016; accepted 4 March 2016
Available online 21 June 2016

KEYWORDS
Child;
Colitis;
Ulcerative/complications;
Tracheitis/etiology

Abstract
Introduction: Inflammatory bowel disease may cause both intestinal and extraintestinal manifestations. Respiratory symptoms in ulcerative colitis are rare and tracheal involvement is exceedingly rare in children.
Case 1: Sixteen year-old female with a 4-week-complaint of abdominal pain, bloody diarrhea, fever and cough. The investigation was consistent with the diagnosis of concomitant ulcerative colitis/coinfection to *Escherichia coli*. On day 4 respiratory signs persisted so azithromycin and inhaled corticosteroids were added. By day 6 she progressed to respiratory failure and was diagnosed with necrotic tracheitis so started on intravenous steroids with fast clinical improvement.
Case 2: Twelve-year-old male adolescent with ulcerative colitis and sclerosing cholangitis started dry cough and throat pain 10 days after diagnosis. Laboratory investigations showed increased inflammatory signs and normal chest X-ray. He started treatment with azithromycin without clinical improvement and on day five he presented dyspnea and fever. Laryngeal fibroscopy suggested tracheitis and so systemic steroids where added with fast clinical and analytic improvement.

* Corresponding author.
E-mail address: isabelserranunes@gmail.com (I.S. Nunes).

http://dx.doi.org/10.1016/j.jpge.2016.03.002
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Discussion: Tracheitis should be suspected if there are persistent respiratory symptoms even when exams are normal. Early recognition and early treatment are essential for a good prognosis preventing progression to respiratory failure.
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PALAVRAS-CHAVE
Criança;
Colite;
Ulcerosa/complicações;
Traqueíte/etiologia

Traqueíte – Uma Complicação Rara de Colite Ulcerosa em Crianças

Resumen
Introdução: A doença inflamatória intestinal pode ser causa de complicações intestinais e extraintestinais. As manifestações respiratórias de colite ulcerosa são raras e o envolvimento traqueal é extremamente raro em crianças.
Caso 1: Adolescente do sexo feminino, de dezasseis anos de idade, com queixas de dor abdominal, diarreia sanguinolenta, febre e tosse seca com 4 semanas de evolução. A investigação realizada foi compatível com o diagnóstico de colite ulcerosa/co-infecção a *Escherichia coli*. No 4º dia de internamento, por persistência das queixas respiratórias, iniciou azitromicina e corticoterapia inalada. Dois dias mais tarde evoluiu para insuficiência respiratória; foi-lhe diagnosticada traqueíte necrotizante pelo que iniciou corticóides endovenosos com rápida melhora clínica.
Caso 2: Adolescente do sexo masculino de 12 anos de idade com colite ulcerosa e colangite esclerosante que inicia queixas de odinofagia e tosse seca 10 dias após o diagnóstico. O estudo analítico revelou aumento dos parâmetros inflamatórios e radiografia de tórax normal. Nesta altura inicia azitromicina, sem melhora clínica, iniciando, no quinto dia de doença, dispnéia e febre. A fibroscopia laríngea foi sugestiva de traqueíte pelo que iniciou corticoterapia sistémica com rápida melhoria clínica e analítica.
Discussão: A traqueíte é uma entidade que deve ser equacionada em doentes com doença inflamatória intestinal na presença de sintomas respiratórios persistentes, mesmo com estudo complementar normal. O seu reconhecimento e tratamento precoces são essenciais para um prognóstico favorável e prevenção da progressão para insuficiência respiratória.
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1. Introduction

Inflammatory bowel disease (IBD) has long been recognized to cause both intestinal and extra-intestinal complications. Airway involvement in IBD can affect any part of the respiratory system but the exact incidence and prevalence of this manifestation are not known.[1–4] Sporadic reports of tracheal involvement in ulcerative colitis are exceedingly rare, and are reported in 16 previous adult cases.[5–14] The aim of this paper is to document the occurrence of this rare situation in children and stress the importance of an early diagnosis and treatment of a potential life threatening condition.

2. Clinical cases

2.1. Case 1

A previously healthy 16-year-old female presented with abdominal pain, bloody diarrhea, fever and productive cough for 4 weeks. Physical evaluation showed a normal lung exam and diffuse abdominal pain without tenderness or other peritoneal signs. Laboratory investigations revealed normocytic anemia (hemoglobin 9.5 g/dL, reference value 12–16 g/dL), thrombocytosis (platelets 425,000/μL, reference value 150,000–300,000/μL), increased C reactive protein (CRP) 203 mg/L (reference value <3 mg/L) and negative pANCA; *Escherichia coli* O157 H7 positive stool culture and normal abdominal X-ray. Suspecting infectious colitis she started intravenous metronidazole and ciprofloxacin with no clinical improvement of diarrhea after a week. Subsequent endoscopic investigation and histology were consistent with the diagnosis of ulcerative colitis. During this period she maintained persistent productive cough sometimes spasmodic. Presumptive diagnosis of ulcerative colitis with concomitant infective colitis and respiratory infection was done and she started treatment with messalazin, azithromycin and inhaled corticosteroid. Bacteriology, virology, polymerase chain reaction (PCR) for *Bordetella pertussis* in sputum and serological tests for *Mycoplasma pneumoniae* were negative. By day six she progressed to respiratory failure needing endotracheal intubation and mechanical ventilation. Abundant tracheal secretions were seen and cytological evaluation showed

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